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MALIGNANT TUMOURS OF THE BUTTOCKS

A SURVEY OF 46 CASES

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MALIGNANT tumours of the buttock area, either primary or secondary in type, are uncommon. The following paper is based on the study of 46 such tumours which have been registered with the Clinical and Pathological Register of the Merseyside Cancer Control Organization during an eight-year period: they form 0·1 per cent of all malignant tumours registered during this time. Cases in which a gluteal tumour developed during the course of a previously diagnosed neoplastic process are not included in this series.

The tumours can be roughly divided into five groups, listed below in order of frequency of occurrence:—

1. PRIMARY MALIGNANT TUMOURS OF BONE (*Table I*).—This group can be subdivided into:—
   - *a. Sarcomas*, including those arising from:—
     - i. Non-osteoblastic connective tissue—the fibrosarcomas and pleomorphic sarcomas.
     - ii. Osteoblastic connective tissue—osteogenic sarcomas.
     - iii. Cartilage-forming connective tissue—chondrosarcomas.
   - *b. Chordomas* arising from notochordal remnants.

2. PRIMARY MALIGNANT TUMOURS OF THE EXTRA-OSSEOUS CONNECTIVE TISSUES (*Table II*).—This group included fibrosarcoma, liposarcoma, myxosarcoma, and spindle- and pleomorphic-cell sarcomas.

3. METASTATIC TUMOURS (*Table III*).—Further subdivided into those:—
   - *a.* Occurring within the pelvic bones.
   - *b.* Occurring in the soft tissues.

4. PRIMARY MALIGNANT TUMOURS OF SKIN (*Table IV*).

5. MISCELLANEOUS TUMOURS (*Table V*) in which the identity of the tumour was in doubt. These groups will now be considered in greater detail.

1. PRIMARY MALIGNANT TUMOURS OF BONE

(*Table I*)

a. Sarcomas.—There were 20 cases in this group, of whom 19 were males; they formed 20 per cent of the total number of bone sarcomas registered during the period and were commoner in the older age groups, only 4 patients being under the age of 40 years. The bone of origin was the ilium in 15 cases, the ischium in 4, and the sacrum in 1 case.

With the exception of 1 patient this group presented with pain, which was localized to the buttock and/or of sciatic distribution. Swelling of the buttock was complained of by 4 patients, 1 of whom attributed the swelling to trauma. The survival period tended to be longer in the patients who complained of swelling, 3 of whom survived for longer than 12 months from the onset of symptoms. The interval between the onset of symptoms and the seeking of medical advice was usually less than 6 months, although 4 patients had had symptoms for over 12 months.

Associated bone disease was present in 7 cases. The predisposition of patients with Paget's disease of bone to develop bone sarcoma is well known, and in the present series Paget's disease of bone was present in 5 patients, in 4 of whom it was widespread. One patient showed evidence of healed rickets; and multiple exostoses of the knee and shoulder region were present in the remaining patient, who subsequently developed a large chondrosarcoma of the ilium. Weber in 1866 described the development of a chondrosarcoma in association with multiple exostoses, and more recently Jaffe (1943) in a series of 28 cases of multiple exostoses found 3 cases of chondrosarcoma; he considers that this does not represent the full incidence of malignancy, as many of his patients are children and the sarcomatous change usually supervenes in adult life.

A definite history of trauma to the region was given by 5 patients, 1 of whom had been treated for a prolapsed intervertebral disk, five years before a recurrence of his sciatic pain led to the diagnosis of a chondrosarcoma arising from the ilium. It is of course possible in this case that the chondrosarcoma was of long standing and that it had been responsible for his initial attack of sciatic pain. A patient who developed an osteogenic sarcoma of the ilium had injured his back and sustained a fracture to a lumbar vertebra several years previously. Two other patients who developed osteogenic sarcomas and 1 who developed a chondrosarcoma all attributed their symptoms to severe falls.

Table I.—PRIMARY MALIGNANT TUMOURS OF BONE

CASE	AGE	SEX	BONE	BONE DISEASE OR TRAUMA	SYMPTOMS	INITIAL DIAGNOSIS	TUMOUR TYPE	TREATMENT	SURVIVAL FROM ONSET OF SYMPTOMS
a. Sarcomas:— D. B.	16	M.	Ischium	Trauma to area preceded symptoms	Pain in hip	Osteochondritis	Chondrosarcoma (H)	Palliative H.V.X.	1 year. Metastasis to thoracic cavity
M. F.	23	M.	Ilium	Multiple exostoses	Tumour in buttock	Sarcoma	Chondrosarcoma (H)	Excision	3 years. Local recurrence
E. H.	40	M.	Ilium spreading to pubes	Prolapsed disk	Sciatica	Recurrence of disk lesion	Chondrosarcoma (H)	Symptomatic	5 years. Metastasis to thoracic cavity
H. R.	21	M.	Ilium spreading to ischium and sacrum	—	Sciatica	Chronic infective arthritis	Osteogenic sarcoma	Palliative H.V.X.	11 months
E. J. W.	48	M.	Ilium spreading to ischium and pubes	Injury to back, fracture lumbar spine	Sciatica	Result of previous injury	Osteogenic sarcoma	Palliative H.V.X.	4 years. Metastasis to thoracic cavity
R. J. J.	49	M.	Ilium spreading to sacrum	Fall preceded symptoms	Painful lump in buttock	Sarcoma	Osteogenic sarcoma	Palliative H.V.X.	1 year
A. E. R.	61	M.	Ilium	Paget's disease, fall preceded symptoms	Pain in buttock	Sarcoma	Osteogenic sarcoma	Radical H.V.X.	8 months
P. M. D.	65	M.	Ilium-pubes	—	Pain in buttocks and hip	Sarcoma	Osteogenic sarcoma	Palliative H.V.X.	2 months
W. E. L.	68	M.	Sacrum	—	Paresis, pain and swelling right leg	Sarcoma	Osteogenic sarcoma	Symptomatic	8 months
T. H.	72	M.	Ilium	Rickets	Sciatica and pain in buttock	Sarcoma	Osteogenic sarcoma (H)	Symptomatic	1 year
G. B.	20	M.	Ilium, ischium, and pubes	—	Pain in buttock	Ischiorectal abscess	Pleomorphic sarcoma (H)	Palliative H.V.X.	9 months
R. W. H.	63	M.	Ilium	—	Pain in hip and buttock	Sarcoma	Pleomorphic sarcoma (H)	Symptomatic	14 months
E. B.	52	M.	Ilium	Paget's disease	Pain in buttock and sciatica	Sarcoma	Fibrosarcoma (H)	Palliative H.V.X.	7 months. Metastasis in thoracic cavity
G. K.	53	M.	Ischium	Paget's disease	Pain in buttock and sciatica	Sarcoma	Fibrosarcoma (H)	Palliative H.V.X.	15 months
W. D.	55	M.	Ischium	—	Pain in hip	Arthritis	Pleomorphic fibrosarcoma (H)	Hind-quarter amputation	11 months
H. S.	60	M.	Ilium	—	Sciatica	Sarcoma	Fibrosarcoma (H)	Palliative H.V.X.	3½ years
R. H.	71	M.	Ilium	Paget's disease	Pain in buttock	Perinephric abscess	Not verified	Testosterone	6 months
W. J. A.	63	M.	Ilium	Paget's disease	Sciatica and swelling	Sarcoma	Not verified	Several courses of H.V.X.	Alive 8½ years
A. E.	69	F.	Ilium	—	Painful swelling of buttock	Sarcoma	P.M. sarcoma of ilium	—	Few months
H. P.	45	M.	Ischium	—	Sudden onset of sciatica	Sarcoma	Not verified	Symptomatic	4 months
b. Chordomas:— S. C.	65	M.	Sacrum	—	Pain in sacral region	—	Chordoma	H.V.X. + surgery	3 years
M. S.	72	F.	Sacrum	—	Pain in sacral region	—	Chordoma	Surgery + H.V.X.	3 years
W. R. H.	72	M.	Sacrum	—	Pain in sacral region	—	Chordoma	Surgery + H.V.X.	4 years

Diagnosis in the older age groups as a rule presented little difficulty, as a tumour was usually obvious on clinical examination; however, in 1 patient the tumour was initially considered to be a perinephric abscess. A recurrence of sciatic pain in the 2 patients who had previously been treated for traumatic bone lesions—i.e., fractured lumbar spine and prolapsed intervertebral disk—was initially attributed to these causes.

Diagnosis was more difficult in the younger patients, in 3 of whom the correct diagnosis was not established for several months, provisional diagnoses of ischiorectal abscess, osteochondritis, and chronic infective arthritis being made; the lesions were in fact pleomorphic sarcoma of the ilium, chondrosarcoma of the ischium, and osteogenic sarcoma of the ilium respectively.

The tumours all attained a large size, the largest being about 15 cm. diameter. They spread into the adjacent bones (*Table I*) and into the neighbouring connective tissues and muscles, forming large irregular tumour masses, stretching, and in some cases ulcerating, the overlying skin. Extension also occurred inwards into the pelvic cavity.

The prognosis of this group of sarcomas was poor and the survival time was as a rule a matter of months, only 4 patients surviving longer than twelve months after seeking medical advice.

Although the series is small, it follows the expected lines in that (*a*) the longest survival time could be expected in the patients with chondrosarcomas, all of whom survived longer than twelve months from the onset of symptoms, and (*b*) the survival time was short in patients in whom a sarcomatous change arose in association with Paget's disease; in the present series death occurred in 3 of these 5 patients within eight months from the onset of symptoms. There did not appear to be any significant difference in the survival time between the osteogenic and the non-osteogenic sarcomas, the time varying in each group between a few months and several years from the onset of symptoms. The longest survival time in the series is in a patient who is still alive 8½ years after the initial diagnosis of Paget's disease complicated by a sarcoma of the ilium. He has had several courses of deep radiotherapy to the original tumour and to local recurrences. Unfortunately histological verification of this case is lacking.

Metastatic growth was diagnosed in 4 patients (*Table I*), in each of whom intrathoracic metastasis

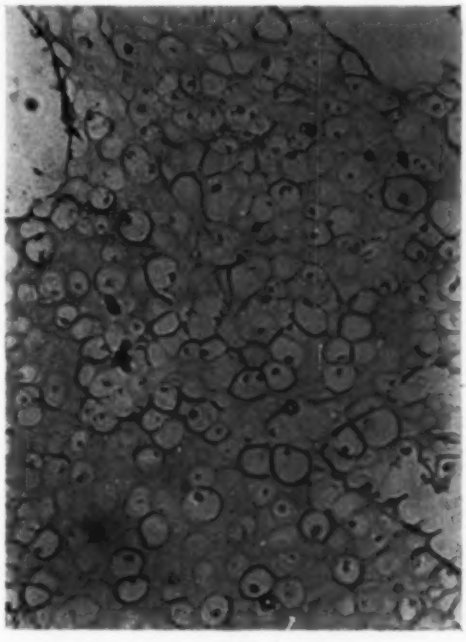

FIG. 129.—*Case* M. F. Chondrosarcoma of the ilium. (× 250.)

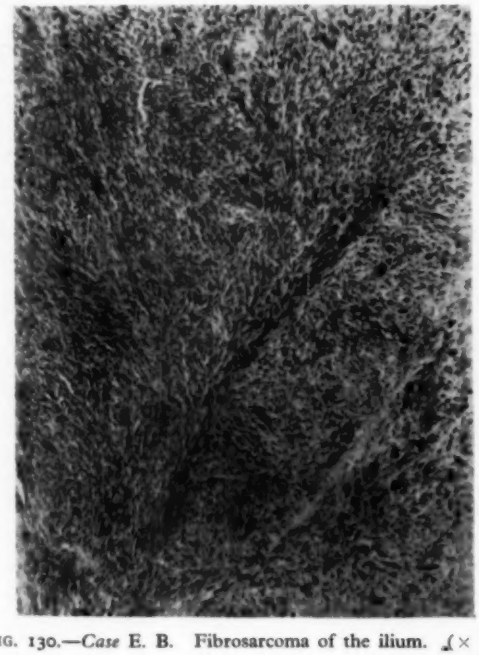

FIG. 130.—*Case* E. B. Fibrosarcoma of the ilium. (× 50.)

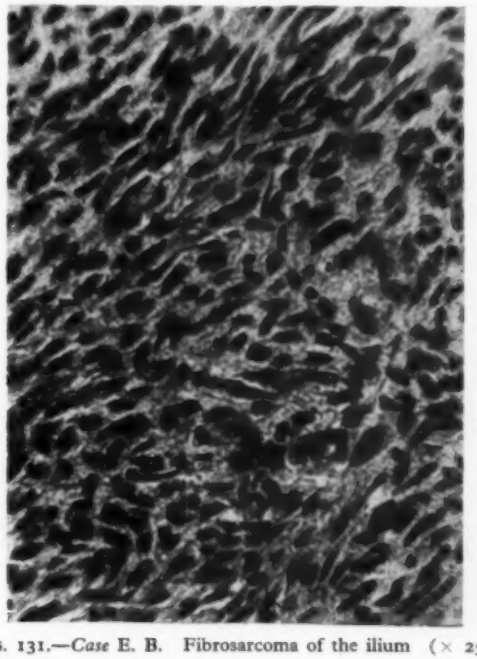

FIG. 131.—*Case* E. B. Fibrosarcoma of the ilium (× 250.)

occurred. This is a somewhat lower incidence of metastasis than one would expect, particularly in regard to osteogenic sarcoma; however, several of the patients died at home and it is possible that metastatic deposits were present but unrecognized.

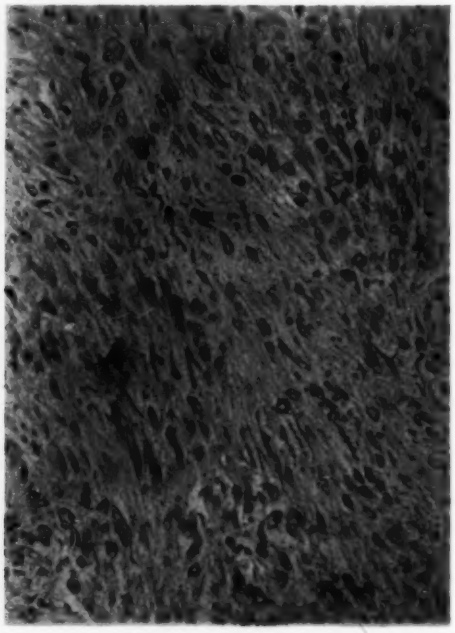

FIG. 132.—*Case* G. B. Pleomorphic sarcoma of the ilium. (× 250.)

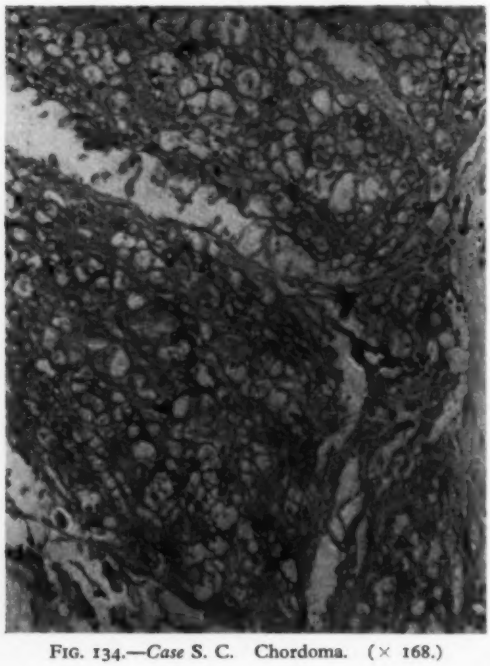

FIG. 134.—*Case* S. C. Chordoma. (× 168.)

Histological examination of the tumour was carried out in 10 cases (*see Table I*) and the histological types included 3 examples of chondrosarcoma (*Fig.* 129), 4 of fibrosarcoma (*Figs.* 130, 131), 2 of pleomorphic sarcoma (*Fig.* 132), and 1 of osteogenic sarcoma (*Fig.* 133). Of the remaining 10 cases there were 6 in which the radiological and clinical features were those of osteogenic sarcoma, and 4 which were soft-tissue sarcomas.

Apart from the expectation of a longer survival time in the cases of chondrosarcoma and the shorter survival time when the sarcoma was a complication

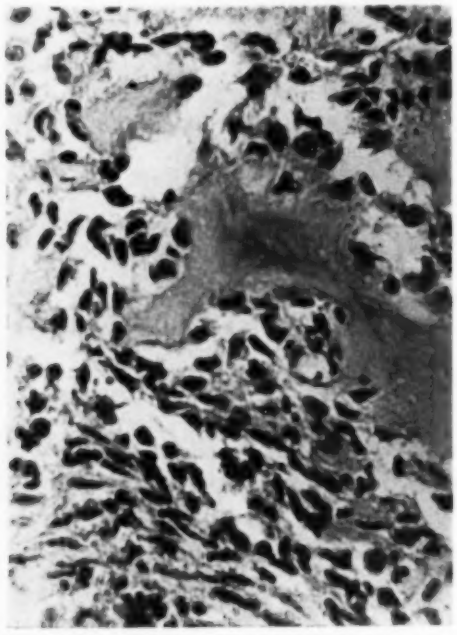

FIG. 133.—*Case* T. H. Osteogenic sarcoma of the ilium. (× 250.)

of Paget's disease, mentioned above, there did not appear to be any correlation between the histological type of the tumour and the clinical picture.

In this series radical removal of the tumour was attempted on two occasions; however, one patient died shortly after the operation, and in the second a recurrence of the tumour—a chondrosarcoma—occurred within a few months. Twelve patients received palliative radiotherapy with a temporary relief of pain, and in some cases also a regression of tumour size.

b. Chordoma.—The first description of a sacrococcygeal chordoma was given by Feldman (1910); since this time descriptions of their histological appearances have been given by many authors including Stewart (1922) and Cappell (1928), and although the tumours are uncommon, surveys of over 100 cases have been culled from the literature by Mabrey (1935) and Gentil and Coley (1948).

There were 3 examples of chordoma in this series. They occurred in elderly patients, 2 of whom were males. Pain in the lower back of 2 to 3 years' duration, resulting from pressure of the tumour on the spinal nerve-roots, was the presenting symptom in each case. Biopsy examination was followed by radical excision of the tumours, and in 2 of the patients palliative radiotherapy for the relief of pain. The ultimate prognosis in this series was poor; in spite of the apparently complete removal of the tumour death occurred between 3 and 4 years after the onset of symptoms.

Histological examination of the gelatinous, lobulated tumours showed the typical appearances of

chordoma (*Fig.* 134), with groups of vacuolated cells, separated by a mucoid matrix and intersected by fibrous trabeculæ.

2. PRIMARY MALIGNANT TUMOURS OF THE EXTRA-OSSEOUS CONNECTIVE TISSUES

(*Table II*)

There were 10 cases in this group, varying in age from 15 to 84 years, and, in contrast to the sarcomas arising from the pelvic bones, the condition was commoner in females (6 cases) than in males (4 cases), and the commonest presenting symptom was unilateral swelling of the buttock, followed in 2 cases by pain, in 1 of these 2 cases the onset of symptoms coinciding with trauma to the area. Pain was the presenting symptom in only 1 case.

The interval between the onset of symptoms and the seeking of medical advice tended to be longer than in the previous group. In 7 cases it was over 12 months; however, in the remaining 3 patients it was only a few months.

An attempt at radical excision of the tumour was made in each case. The excised tumours varied in size between 5 cm. and 25 cm. in their longest axis, and were lying apparently encapsulated between the gluteal muscles. Two of the largest tumours extended down as far as the periosteum overlying the ilium, and in one of these ulceration of the overlying skin was present at the initial examination. Apart from the apparent encapsulation the tumours did not present any characteristic macroscopical appearances, varying in consistency between soft cystic masses and firm tumours, and presenting a smooth or knobbly exterior.

The tissue of origin of this group of tumours was difficult to ascertain, probably it was the intermuscular connective tissue or muscle-sheaths. Histological examination was carried out in each case; there were 4 examples of fibrosarcoma, 2 of myxosarcomas (*Fig.* 135); the remaining cases were examples of spindle-cell sarcoma, mesonephroma (*Fig.* 136), pleomorphic sarcoma, and liposarcoma.

Table II.—SOFT-TISSUE SARCOMAS

CASE	AGE	SEX	TRAUMA	SYMPTOMS	HISTOLOGICAL APPEARANCES	TREATMENT	SURVIVAL FROM ONSET OF SYMPTOMS
F. S.	15	M.	Nil	Swelling 2 months	Fibrosarcoma	Palliative H.V.X.	12 months. Local recurrence
F. J.	25	M.	Nil	Swelling and pain 2 months	Myxosarcoma	Radical surgery to tumour and recurrence	13 months. Local recurrence
B. L.	40	F.	Nil	Swelling 2 years; recently more rapid growth and pain	Spindle-cell sarcoma	Surgery	5 years. Local recurrence + secondaries in lung, alimentary tract, and bone
J. R. B.	45	M.	Trauma preceded symptoms	Swelling buttock 3 years, sciatica 1 year	Myxosarcoma	H.V.X. and surgery	7 years. Secondaries in omentum and lung. Primary well
E. J.	55	F.	Nil	Pain in back and thigh 2 years	Fibrosarcoma	Palliative H.V.X.	2½ years. Local recurrence
E. M.	58	F.	Nil	Recurrent gluteal swellings	Myxoma-mesonephroma	Surgical excision of tumour and recurrences	17 years. Local recurrences and secondaries in abdomen
E. P.	63	F.	Nil	Recurrent gluteal swellings	Pleomorphic sarcoma	Surgical excision of tumour and recurrences + palliative H.V.X.	5 years. Local recurrences
G. J. G.	70	F.	Nil	30-year history 'pimple'; 2 years rapid growth	Fibrosarcoma	Surgery	Alive and well 5 years
S. A.	72	F.	Nil	Recurrent gluteal swellings	Lipoma-sarcoma	Surgery and H.V.X.	Several years. Secondaries in lung, primary well
R. S.	84	M.	Nil	Lump in buttock 5 months	Fibrosarcoma	Surgery	12 months

An interesting feature of this group of tumour was its tendency to local recurrence; in 4 of the cases the growth recurred on several occasions. One patient was symptom-free for 12 years after an excision of a 'myxoma' of the buttock, but in the succeeding 5 years recurrences of the tumour were removed on four occasions, the histological picture being that of a mesonephroma. At her death secondary deposits were present in the abdominal cavity. In 2 other cases the tumours recurred on two occasions, in 1 of these cases the original tumour had the appearances of a lipoma, the recurrent growths having undergone a sarcomatous change: in the other, the original tumour was a pleomorphic sarcoma. In the fourth patient a local removal of a myxosarcoma was followed 12 months later by a hindquarter amputation for a recurrence.

The survival time of this group was of longer duration than in the previous group of sarcomas; all of the patients survived 12 months from the

onset of their symptoms, and in 7 the survival period varied between 2 and 17 years. It is interesting to note that the shortest survival periods were in the two youngest and in the eldest patient. Metastatic deposits were present at the time of death in 4 cases, in 2 of these the histological and clinical findings were suggestive of a bronchial origin.

With the exception of 1 patient pain was the presenting symptom, usually localized to the buttock area. The patients usually sought advice within a

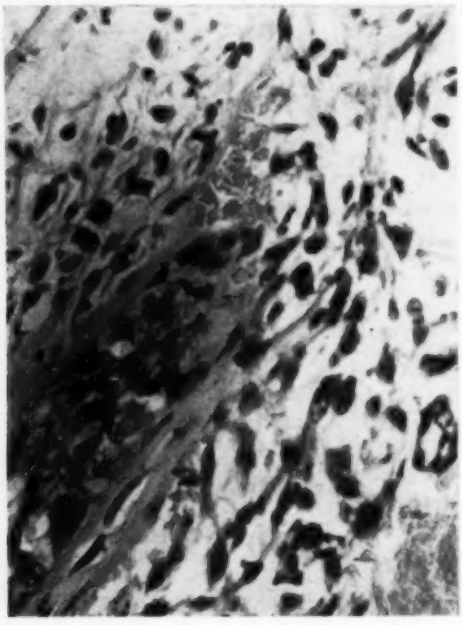

FIG. 135.—*Case* J. R. R. Myxosarcoma of the soft tissues. (× 250.)

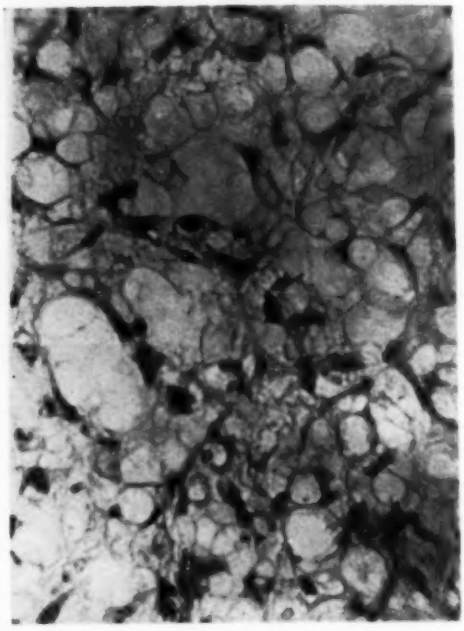

FIG. 136.—*Case* E. M. Mesonephroma of the soft tissues. (× 250.)

in 2 of whom there was no residual tumour in the buttock. In the remaining 7 cases large local recurrences were also present.

3. METASTATIC TUMOURS

(*Table III*)

Carcinomatous metastasis to the skeletal system is common—Abrams, Spiro, and Goldstein (1950) found an incidence of 27 per cent in a series of 1000 autopsies on cancer patients; however, only a small percentage of these growths manifest themselves clinically, either through the development of pain, a pathological fracture, a palpable tumour, or involvement of the spinal cord. The predilection of carcinoma metastases to the innominate bones is well known, but it is rare for a metastatic growth in this site to be the first manifestation of the patient's neoplastic condition.

Blood-borne metastatic deposits in voluntary muscle are rare, although they have been reported from several primary sites including bronchus.

In the present series of cases there were 3 patients in whom the initial symptoms were due to a solitary metastatic growth in the innominate bone, and 3 in whom the initial symptoms were due to a solitary metastatic growth in the gluteal muscles, with secondary involvement of the underlying bone in 1 case. These 6 patients belong to the older age groups, and 5 were males. The primary growth was subsequently located in the bronchus on three occasions. In the other 3 cases the site of the primary growth was not definitely located, although few months of the onset of symptoms, although 1 had complained of 'sciatica' for 9 months before noticing a lump in the sacro-iliac region, for which she sought advice; another gave a history of having fallen down a manhole 3 years previously—this was followed by throbbing pain in his thigh which had steadily become worse and shortly before attending hospital he had noticed a swelling of the corresponding buttock.

Histological examination of all the tumours was carried out, in 3 cases on operative specimens, in 2 on post-mortem specimens, and in the last case both operative and post-mortem material was available.

Of the patients in whom the primary growth was subsequently located in the bronchus: the first attended hospital with a 12 months' history of sciatic pain. Clinical examination revealed a gluteal mass posterior and exterior to the ilium, extending into the perineum, and backwards to the sacro-iliac joint. Radiological examination showed a soft-tissue mass arising from, and destroying, the ischium, with appearances suggestive of an osteogenic sarcoma. There was at this time no evidence of any pulmonary pathology. The patient died 6 months later and post-mortem examination showed a white fleshy growth at the hilum of the left lung, with metastatic deposits in the hilar and abdominal lymph-nodes, both suprarenal glands, and a soft bony tumour arising from the ramus of the pubis, extending along the inner aspect of the pelvis to the sacro-iliac joint. Histological examination of the various tumours showed a well-differentiated adenocarcinoma (*Fig.* 137).

Table III.—Metastatic Tumours

Case	Age	Sex	Trauma	Initial Symptoms	Clinical and Radiological Impressions	Histological Appearances	Treatment	Primary Site	Survival from Onset of Symptoms
H. A.	45	M.	Nil	Pain and swelling of buttock	Radiologically an osteogenic sarcoma of pelvis	Adenocarcinoma	Palliative H.V.X.	P.M. Carcinoma of bronchus; secondaries in mediastinal glands, suprarenals, and bones	18 months
P. H.	60	M.	Nil	Pain and swelling of buttock	Tumour	Anaplastic carcinoma	Local excision	Bronchus. P.M. Carcinoma right bronchus + secondaries in brain, skin, bones, and lymph-glands	6 months
W. E. J.	63	M.	Nil	Pain in buttock	Swelling of buttock. Radiologically a chondrosarcoma of ilium	Keratinizing squamous carcinoma	Biopsy. Palliative H.V.X.	?	Dead 1 year local recurrence + chest symptoms
M. McL.	66	F.	Nil	Pain in buttock followed by tumour	Tumour of buttock	First excision, keratinizing squamous carcinoma. Second excision, oat-cell carcinoma	Attempt at radical excision; post-operative irradiation	Probably bronchus	2 years. Local recurrence + metastatic growth in cervical glands and lung
G. P.	77	M.	Trauma preceded symptoms	3-year history of tenderness of buttock. Tumour of more recent onset	Radiologically secondary deposit in ilium	P.M. anaplastic carcinoma	Symptomatic	Bronchus. Secondary deposits also in lymph-glands and brain	6 months since swelling noted
J. C.	85	M.	Nil	Bleeding P.R. and swelling of buttock	Ischiorectal abscess	Papillary carcinoma	Excision	?	Alive at 1½ years

The second patient gave a history of pain in his left hip of 3 years' duration and a gluteal swelling of more recent onset. On examination there was a large tender mass extending backwards from the anterior iliac spine into the gluteal region. Radiological appearances of the tumour were those of a metastatic deposit, and a radiograph of the chest was very suggestive of there being a bronchial neoplasm.

Fig. 137.—*Case* H. A. Secondary adenocarcinoma. Primary growth located in bronchus. (× 250.)

The patient died 3 months later and post-mortem examination showed a carcinoma of the bronchus, with secondary deposits in lymph-nodes, and the innominate bone; death was due to a hæmorrhage into a metastatic cerebral growth. Histological examination of the tumour showed a very heterogeneous appearance, varying from a poorly differentiated squamous carcinoma, with prickle cells, cell nests, and keratin production (*Fig.* 138), to areas of frank colloid carcinoma with abundant extracellular mucus and typical signet-ring cells (*Fig.* 139) and large areas of necrosis.

The third patient attended hospital with a history of pain in his buttock of several months' duration and a tumour of more recent onset. A lobulated tumour about the size of an egg was removed from the soft tissues of the gluteal region. Histological examination showed a spheroidal-cell carcinoma with palisading of the basal nuclei and large areas of necrosis (*Fig.* 140). The patient's condition deteriorated rapidly and post-mortem examination showed a carcinoma of the bronchus to the right upper lobe, with metastatic deposits in the left lung, cervical and mediastinal lymph-nodes, dorsal and lumbar vertebræ, brain, chest wall, and buttock.

In one of the cases in which the site of the primary growth was in doubt the tumour formed a large mass, arising from the left ilium and extending on to the sacrum. The radiological appearances were very suggestive of a chondrosarcoma; however,

histological examination showed a typical, poorly differentiated keratinizing squamous carcinoma (*Fig.* 141). The tumour responded well to radiotherapy, but recurred locally and the patient died rather suddenly with symptoms of pulmonary pathology. Unfortunately a post-mortem examination was not carried out in this patient; however, a radiological examination had revealed a symptomless pleural effusion and it is possible that the primary growth may have been in the bronchial tree.

In the second patient in whom the site of the primary growth was in doubt, the gluteal tumour formed a large mass in the soft tissues of the buttock, spreading down to involve the sacro-iliac joint. This tumour was excised and the histological picture varied from a poorly differentiated squamous carcinoma (*Fig.* 142) with prickle cells, cell nests, and keratin formation to areas of mucoid degeneration with large multinucleate physaliferous cells. This picture was very reminiscent of one of the cases in

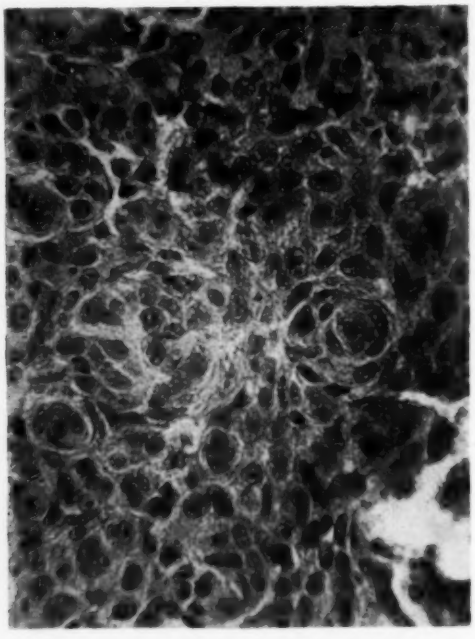

FIG. 138.—*Case* G. P. Secondary squamous carcinoma showing cell nest formation and prickle cells. Primary growth located in bronchus. (× 250.)

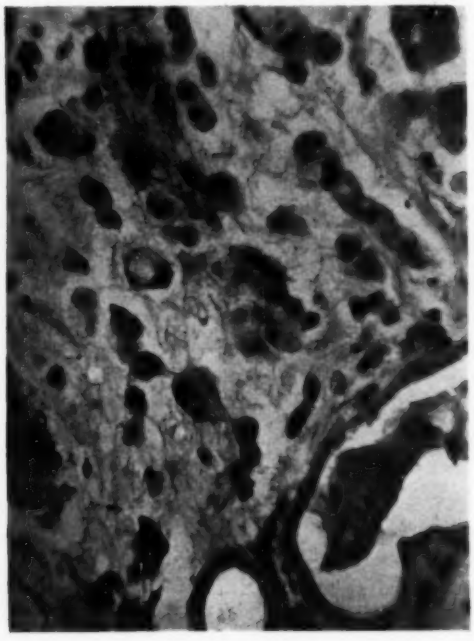

FIG. 139.—The same case as *Fig.* 138 with areas of colloid adenocarcinoma. (× 250.)

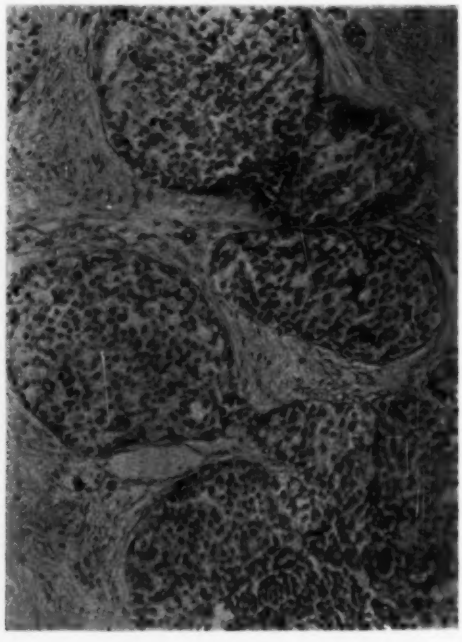

FIG. 140.—*Case* P. H. Secondary spheroidal-cell carcinoma. Primary growth located in bronchus. (× 250.)

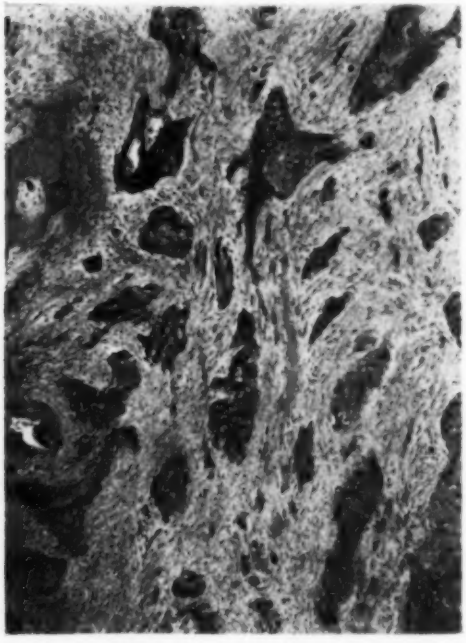

FIG. 141.—*Case* W. E. J. Secondary squamous carcinoma invading the ilium. Primary growth not located. (× 50.)

which the primary growth was definitely located in the bronchus. A recurrence of this tumour was removed six months later and the histological appearances were completely different, being very suggestive of a fibrosarcoma (*Fig.* 143); however, in view of the earlier findings, it was more correctly identified as

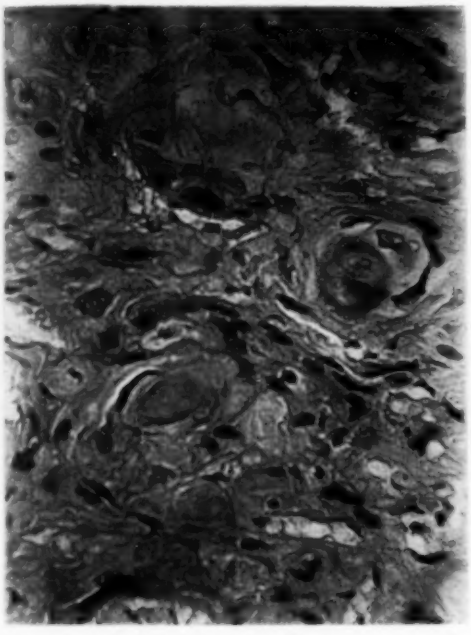

FIG. 142.—*Case* M. McL. Secondary squamous carcinoma. This section was prepared from the first operative specimen. Primary growth not located. (× 250.)

FIG. 144.—*Case* J. C. Papillary adenocarcinoma. (× 50.)

an anaplastic type of oat-cell carcinoma. This patient developed dyspnœa and enlargement of the cervical glands shortly before her death; these symptoms, together with the histological appearances of the gluteal tumour, are again suggestive of the primary growth having been in the bronchial tree.

The remaining patient in this group presented with bleeding per rectum and a gluteal swelling of two weeks' duration, diagnosed initially as an

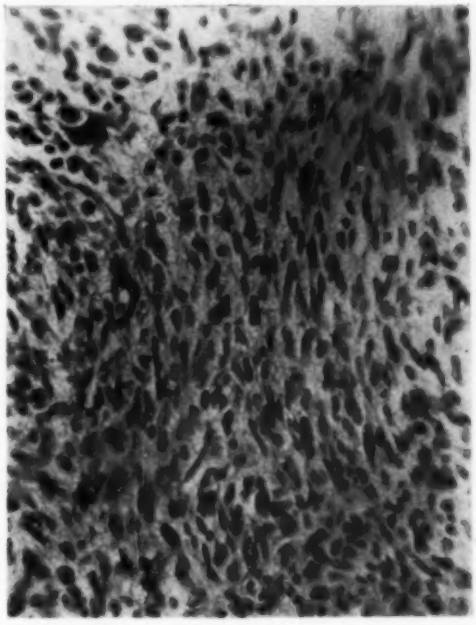

FIG. 143.—Secondary anaplastic carcinoma, simulating fibrosarcoma. Same case as *Fig.* 142. (× 250.)

ischiorectal abscess. Histological examination of the curettings from the abscess showed a papillary adenocarcinoma (*Fig.* 144). Examination of the patient at this time also revealed an obstruction to the right ureter which proved to be a papilliferous type of transitional carcinoma, and dissimilar histologically from the gluteal growth. He is still alive 9 months after his operation. It is possible that this gluteal tumour has arisen from some embryological remnant, and is in fact a primary rather than a secondary growth.

It is interesting to note that whilst the prognosis of this group of tumours is hopeless, they all survived at least 6 months from the onset of symptoms and 4 survived for longer than 12 months.

4. PRIMARY MALIGNANT SKIN TUMOURS

(*Table IV*)

These were uncommon tumours in this site and the 3 cases in this group complained of a 'wart' on the buttock for periods varying from 12 to 30 years. In 2 cases the tumours were squamous carcinomas and in the third a basal-cell carcinoma. Two were treated by radiotherapy and one surgically, and have all been symptom-free since treatment.

5. MISCELLANEOUS TUMOURS

(*Table V*)

Four tumours in young patients are included in this group.

The youngest was a male infant in whom a 'pimple' was noticed on the buttock at three months. He attended hospital a month later, and on examination there was a large hard mass 10 cm. diameter, fixed deeply in the right buttock. A biopsy showed a completely undifferentiated polyhedral-cell carcinoma (*Figs.* 145, 146), tending in some parts to be arranged in columns and in others infiltrating between the muscle-fibres and through the connective tissue. At this time a swelling was also noted in the left lower jaw, which had the radiological appearances of a secondary carcinoma; palliative radiotherapy was given with a good immediate response. However, 2 months later the child died with residual malignancy in the buttock, and metastatic deposits in the mandible.

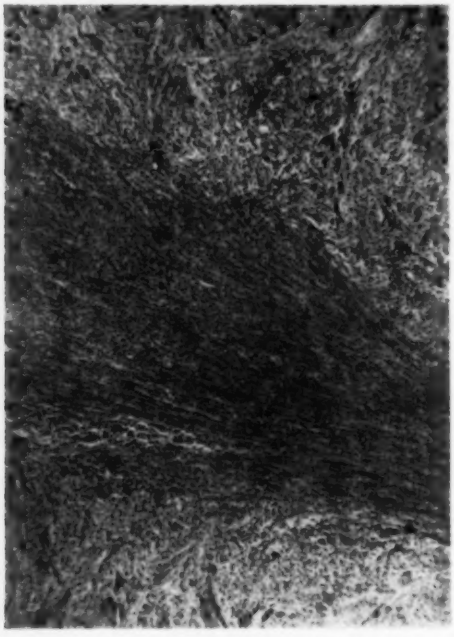

FIG. 145.—*Case* E. A. W. Undifferentiated polyhedral-cell carcinoma infiltrating subcutaneous tissues. (× 50.)

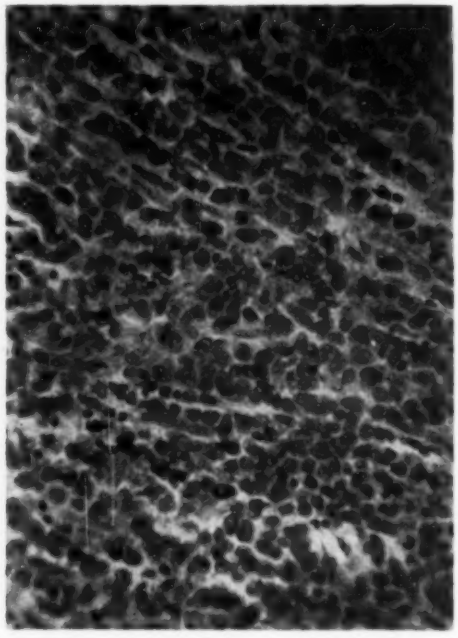

FIG. 146.—Same case as *Fig.* 145 showing cellular detail. (× 250.)

Table IV.—SKIN SARCOMA

CASE	AGE	SEX	SYMPTOMS	HISTOLOGICAL APPEARANCES	TREATMENT	SURVIVAL FROM ONSET OF SYMPTOMS
G. W. D.	30	M.	'Pimple' on buttock	Squamous epithelioma	Diathermy excision	Alive 13 years
E. M.	49	F.	'Wart' on buttock	Basal cell carcinoma	Radiotherapy	Alive 34 years
W. W. L.	70	M.	'Pimple' on buttock	Squamous epithelioma	Radiotherapy	Alive 20 years

Table V.—MISCELLANEOUS TUMOURS

CASE	AGE	SEX	TRAUMA	SYMPTOMS	HISTOLOGICAL APPEARANCES	TREATMENT	SURVIVAL FROM ONSET OF SYMPTOMS
E. A. W.	3 mth.	M.	Nil	Swelling of buttock	Anaplastic carcinoma	Palliative radiotherapy. Good initial response	3 months. Metastatic growth in mandible. Recurrence in buttock
S. W.	2 yr.	F.	Nil	Sacral swelling and pain	? Embryoma	Symptomatic	5 months. Metastatic growth in lungs. Residual tumour in buttock
L. J.	17 yr.	F.	Fall whilst skating	Swelling + pain in ischiorectal fossa	? Anaplastic carcinoma. ? Reticulosis	Palliative radiotherapy. Dramatic initial response	6 months. Generalized spread in lymphatic system. Recurrence in buttock
L. Y. N.	30 yr.	M.	Nil	Hæmatemesis + sciatica	? Reticulosis	Palliative radiotherapy. Good response	Alive 3 years. Metastatic growth in inguinal region. Primary site well

The second case was a female child 2 years of age. She attended hospital with a history of a swelling in the sacral region, associated with pain on sitting and on defæcation, for 2 months. On examination there was a large, tense swelling between the anus and sacrum, extending outwards to the

region of the left buttock, where it was infiltrating the gluteal muscles. Histological examination of a biopsy specimen of the tumour showed a rapidly growing carcinoma consisting of large, polyhedral cells with abundant foamy cytoplasm, the nucleus were ill formed and areas of necrosis and hæmorrhage frequent. The exact nature of the tumour was in doubt, and it was considered to be a type of embryoma. The child died of a terminal pneumonia three months after her first attendance at hospital, with residual

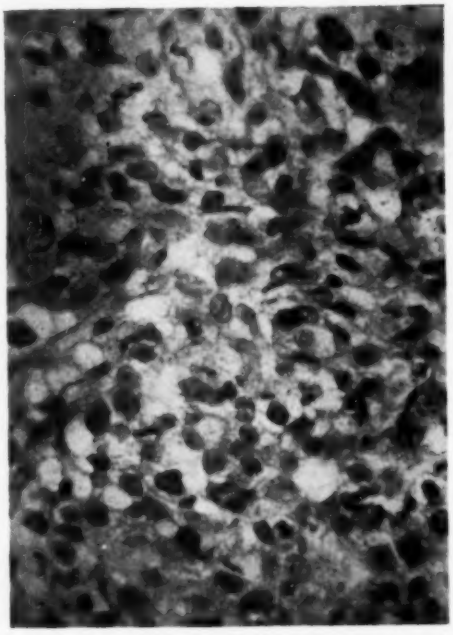

FIG. 147.—*Case* S. W. Section from the tumour considered possibly to be a type of embryoma, showing large, characteristic cells having abundant foamy cytoplasm. (× 268.)

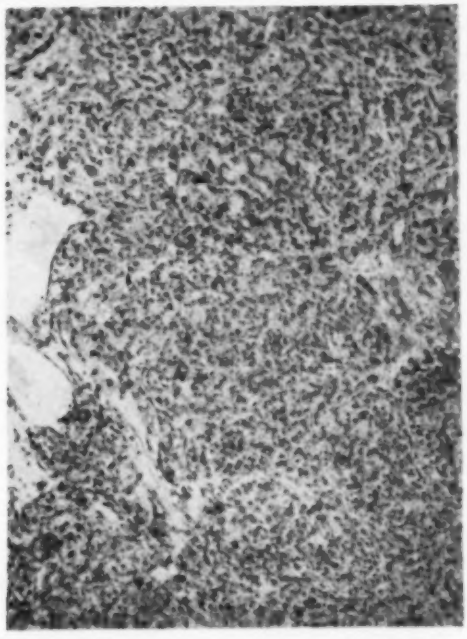

FIG. 148.—Same case as *Fig.* 147, showing sheets of malignant cells, divided by fibrous trabeculæ into solid alveoli. (× 68.)

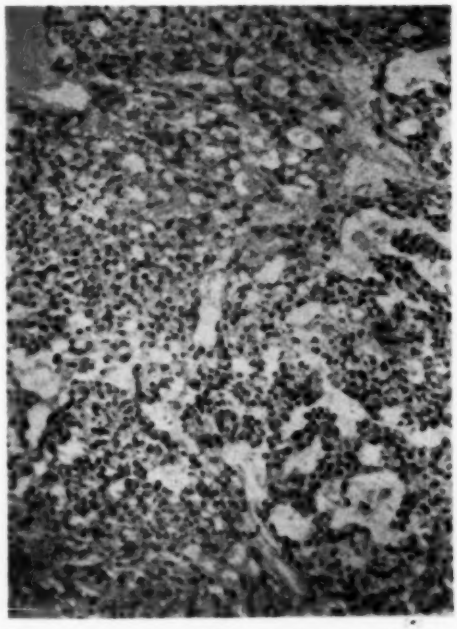

FIG. 149.—Same case as *Figs.* 147 and 148, showing a pseudo-adenomatous arrangement. (× 68.)

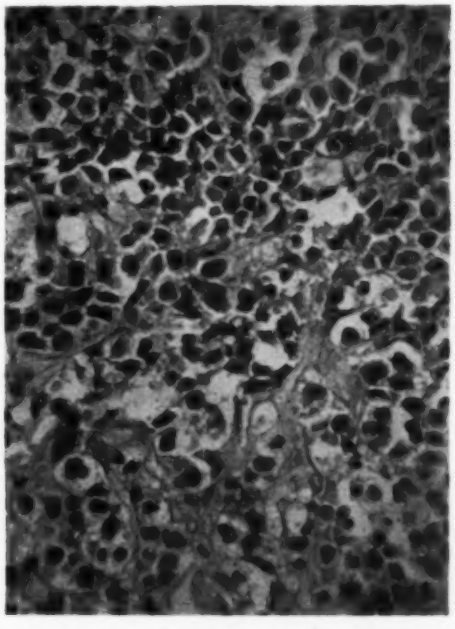

FIG. 150.—*Case* L. J. Anaplastic tumour, possibly a reticulosis. (× 268.)

having a finely reticular pattern and prominent nucleolus (*Fig.* 147). Mitotic figures were numerous. The cells were arranged in some areas as solid alveoli, separated by fibrous trabeculæ (*Fig.* 148), and in others in a cribriform manner, resulting in a pseudo-adenomatous pattern (*Fig.* 149). The blood-vessels tumour in the buttock and with metastatic deposits in the lungs.

The third case was a girl of 17 years who attended hospital with a history of pain and swelling in the ischiorectal fossa following a fall sustained whilst skating. This was diagnosed initially as an ischiorectal

abscess; however, the tumour continued to grow and a month later was a tender, ulcerated mass 10 cm. in diameter, extending down to the ischium, and with metastatic deposits in the inguinal and supraclavicular lymph-glands. A biopsy from the gluteal tumour (*Fig.* 150) showed a rapidly growing anaplastic tumour composed of large cells with a moderate amount of slightly granular cytoplasm and with large vesicular nuclei, some of which contained one or more prominent nucleoli. An occasional multinucleate giant cell was present, and also plasma cells and neutrophil polymorphs. The cells were

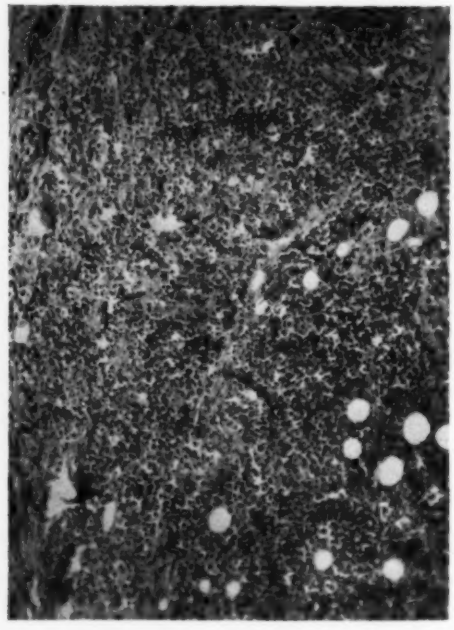

FIG. 151.—Same case as *Fig.* 150, showing sheets of anaplastic cells intersected by fibrous trabeculæ, infiltrating fatty tissue. (× 68.)

infiltrating between muscle bundles, and in some parts were forming large sheets of malignant tissue, separated by fibrous trabeculæ (*Fig.* 151). The nature of the tumour was in doubt—it may have been a completely undifferentiated carcinoma, a type of reticulosis, or possibly it may have arisen from some embryological remnant. The gluteal tumour and glands were treated with palliative radiotherapy, the immediate effect being dramatic. However, the tumour recurred, and the patient died 4 months after the initial treatment with a massive local recurrence of the gluteal tumour, and with generalized lymphatic spread.

The fourth case in this group was a young male aged 30 years. He was admitted to hospital with hæmatemesis which followed the taking of large numbers of aspirin to relieve an attack of 'sciatica'. On examination a tumour of the buttock was noted; it was infiltrating the gluteal muscles and spreading down to the underlying bones. A biopsy of the tumour showed it to be composed of regular, undifferentiated cells, infiltrating the muscle-fibres, and considered at this stage to be a secondary undifferentiated carcinoma from an unknown primary growth. The tumour responded satisfactorily to palliative radiotherapy and the patient remained symptom-free for three years before enlargement of the inguinal glands was noted. The glands were excised, and histologically they showed marked follicular hyperplasia, with focal collections of typical histiocytic cells; these latter were similar to the cells found separating the muscle bundles in the original gluteal tumour. The diagnosis was altered to a variety of a reticulosis.

These 4 cases, all occurring in young patients, and equally divided between the sexes, form a heterogeneous group of tumours of doubtful cytological type and origin. Nevertheless, certain similarities are present amongst them. With the exception of the eldest patient, the tumours were rapidly progressive, and caused death within a few months of the initial symptoms. Metastatic deposits occurred in all cases, in the 2 infants tumour dissemination was via the blood-stream, and in the older 2 patients, in whom the histological appearances were suggestive of a reticulosis, the dissemination was chiefly via the lymphatic system. The 3 tumours treated with palliative radiotherapy all showed a very good initial response, although in 2 of these cases the tumours recurred in a very short time.

DISCUSSION

Malignant tumours of the buttocks receive little attention in either the text-books of surgery or of pathology. A comprehensive article on tumours of the buttocks was written by Charache (1952); he reviewed 64 cases in the literature, of which 13 were malignant, 46 benign, and 5 non-neoplastic, and added a further 30 cases from the records of the Brooklyn Cancer Institute—15 of these cases were malignant (7 neurogenic sarcomas, 2 squamous carcinomas, 1 basi-squamous carcinoma, 1 intradermal carcinoma, 1 hæmangio-endothelial sarcoma, and 3 metastatic carcinomas), 6 benign, and 9 non-neoplastic. He does not appear to have included the primary malignant tumours of bone in his study. Bowden and Booher (1953), in a paper dealing with the surgical treatment of sarcoma of the buttock, described 10 cases of sarcomas of the soft parts of the buttocks. Goldenberg (1954) reported 2 cases of fibrosarcoma of the buttock following the intramuscular injection of a preparation consisting of penicillin and sesame oil.

The 46 malignant tumours of the buttocks reviewed in the present paper occurred over a wide age range, the youngest being a few months old and the eldest over 80 years of age. The youngest patients were included in the miscellaneous group, 3 out of these 4 patients being under 20 years of age. The percentage of patients under the age of 25 years in the group of sarcomas arising from bone was smaller than might be expected (3 out of 20 cases); this is presumably associated with the fact that this review includes only the sarcomas arising from the pelvic bones, whilst the commonly affected bones in children and young adults are the long bones (Lichtenstein, 1952). Chordomas, metastatic carcinomas, soft-tissue sarcomas, and skin carcinomas occurred in patients past middle age with the exception of 2 cases with soft-tissue sarcomas, both of which were under the age of 25 years; this age distribution is comparable with the series of Charache (1952) and Bowden and Booher (1953).

The two most common presenting symptoms were pain, either in the lower back, or of sciatic distribution, and swelling. Pain, with or without accompanying gluteal swelling, was invariably complained of by those patients in whom neoplastic involvement of the pelvic bones, whether primary or secondary in nature, was present; this included the primary malignant tumours of bone and some of the metastatic carcinomas. When the tumour was confined to the soft tissues, swelling, usually unaccompanied by pain in the initial stages, was the presenting symptom. In the 10 cases of soft-tissue sarcomas described by Bowden and Booher (1953) the presenting symptom was invariably a gluteal mass, usually unaccompanied by pain. Of these 30 cases described by Charache (1952), 18 complained of a tumour unaccompanied by pain, and 12 of a tumour accompanied by pain. He does not specify which of these tumours were malignant.

The duration of symptoms before seeking medical advice was correlated to some extent with the tumour group—it was longest in the primary skin cancers (over 10 years in each case), shortest in the sarcomas of bone and the metastatic carcinomas (frequently less than 6 months), and intermediate in the chordomas and soft-tissue sarcomas (usually between 1 and 2 years). In the series of soft-part sarcomas described by Bowden and Booher (1953) this time interval tended to be shorter, over half of their patients seeking medical advice in less than 4 months from the onset of symptoms.

With the exception of the skin carcinomas the tumours grew to a large size, and large residual or recurrent, ulcerated tumours were present terminally in most of the patients. The propensity of soft-tissue sarcomas to recur is well known and was discussed by Warren and Sommer (1936) and by Cade (1951) from the surgical standpoint. In the present series local recurrences were removed on one or more occasions from 4 of these 10 patients.

Metastatic growth occurred in all of the cases in the miscellaneous group, in 2 of whom the tumour disseminated via the lymphatic system and in 2 via the blood-stream. Widespread metastases were present in 5 out of the 6 patients with secondary carcinoma, dissemination occurring by means of both lymphatic and blood systems in each of these 5 patients. Metastatic growth was recorded in 4 of the patients with soft-tissue sarcoma and in 5 of the patients with sarcomas arising from bone; the metastatic growths were located in the lungs in each case. Secondary growths were not recorded in association with either the skin carcinomas or the chordomas.

Trauma may possibly have some bearing on the aetiology of the sarcomas of bone, a definite history of antecedent trauma to the part being given by 5 out of these 20 patients; it did not appear to be significant in any of the other groups. The predisposition of patients with bone disease, particularly osteitis deformans, to develop bone sarcomas was borne out in this series in which associated bone disease was present in 7 out of 20 patients who had sarcomas of bone.

The prognosis of these tumours was very gloomy, radical cure being effected in the skin carcinomas only. Amongst the other groups of tumours the longest average survival time was in the sarcomas of the soft tissues (4½ years); adequate surgical removal of these tumours resulted in the patients being symptom-free for periods varying between a few months and several years before a local recurrence of the tumour occurred. Bowden and Booher (1953), in their paper dealing with the surgical treatment of these tumours, stress the need for radical excision as the tumour is usually more widespread than is appreciated clinically.

Radical surgical excision combined with radiotherapy was attempted on the 3 patients with chordomas. Local recurrence occurred, however, and the average survival period was about 3 years.

Radical therapy was only attempted on 3 patients with sarcoma of bone, in 2 cases surgically, and in 1 case by radiotherapy; however, it did not appear to alter the progress of the disease. Twelve patients received palliative radiotherapy, with the temporary relief of symptoms. The average survival time in this group was less than 2 years, being longest amongst the cases of chondrosarcoma (average survival time 3 years) and shortest in those patients with an accompanying Paget's disease.

In the group of metastatic carcinomas the average survival time was about 12 months; the methods of treatment included local excision and palliative radiotherapy. The survival time was shortest in the miscellaneous group, 3 of the 4 patients included in this group survived less than 6 months from the onset of symptoms.

SUMMARY

Forty-six malignant tumours of the buttock are reviewed. They can be divided into primary tumours of bone—sarcomas and chordomas; sarcomas of the soft tissues; metastatic carcinomas; primary skin tumours; and a miscellaneous group in which the exact nature of the tumour was in doubt.

The tumours occurred over a wide age range, the youngest patient being a few months old and the eldest over 80 years. The incidence of associated bone disease, principally Paget's disease, and also of trauma were significantly increased in the sarcomas of bone.

Pain, either in the back or of sciatic distribution, was the commonest presenting symptom in cases with involvement of the bony pelvis; gluteal swelling was the presenting symptom when the tumour was chiefly confined to the soft tissues.

With the exception of the skin carcinomas the prognosis was poor, the survival times for the different groups varying between a few months to several years.

I wish to thank Dr. J. S. Fulton for his permission to use the clinical material from the register of malignant disease of the Merseyside region, and also all the pathologists who contribute histological material to the Pathology section of the Register.

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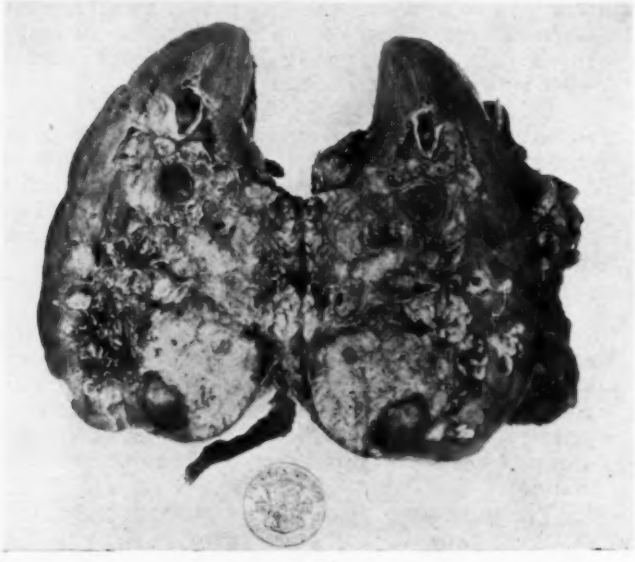
FIVE-YEAR RESULTS OF OPERATION FOR NEPHROMA

USE OF DIAGNOSTIC EXPLORATION IN HÆMATURIA

BY A. WILFRID ADAMS, BRISTOL

THE unusual incidence of ten nephrectomies for neoplasm in adults during one year (1949), led to a publication (Adams, 1950) in which was foreshadowed a further communication giving the sequelæ five years later. The findings are here presented, the cases tabulated in categories according to their fates and retaining the original (chronological) order.

By way of instructive analysis, the events in each individual since 1949 are given following a short extract or precursory history and operation findings. Subsequently, the handling of the bout of hæmaturia is discussed and fallacies of routine investigation resulting at times in dangerous delay of nephrectomy commented on. Recourse to exposure is recommended as the logical answer where diagnosis is unsettled.



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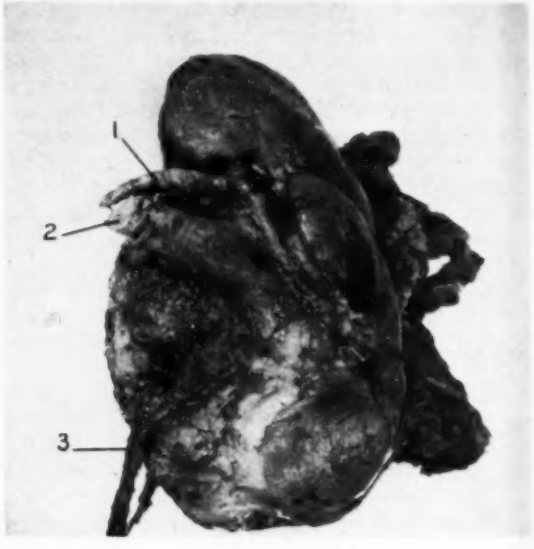

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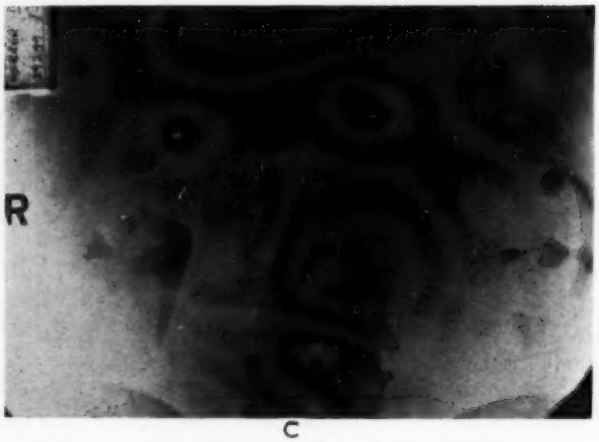


C

FIG. 152.—*Case* W. E. W., male, 52 years. Adenocarcinoma. Massive invasion seen in sectional (A) and external (B) views of kidney (1, Artery ; 2, Vein with growth protruding ; 3, Ureter bent forward by post-medial neoplasm). C, Pyelogram showing calices directed inwards from lateral pelvis.

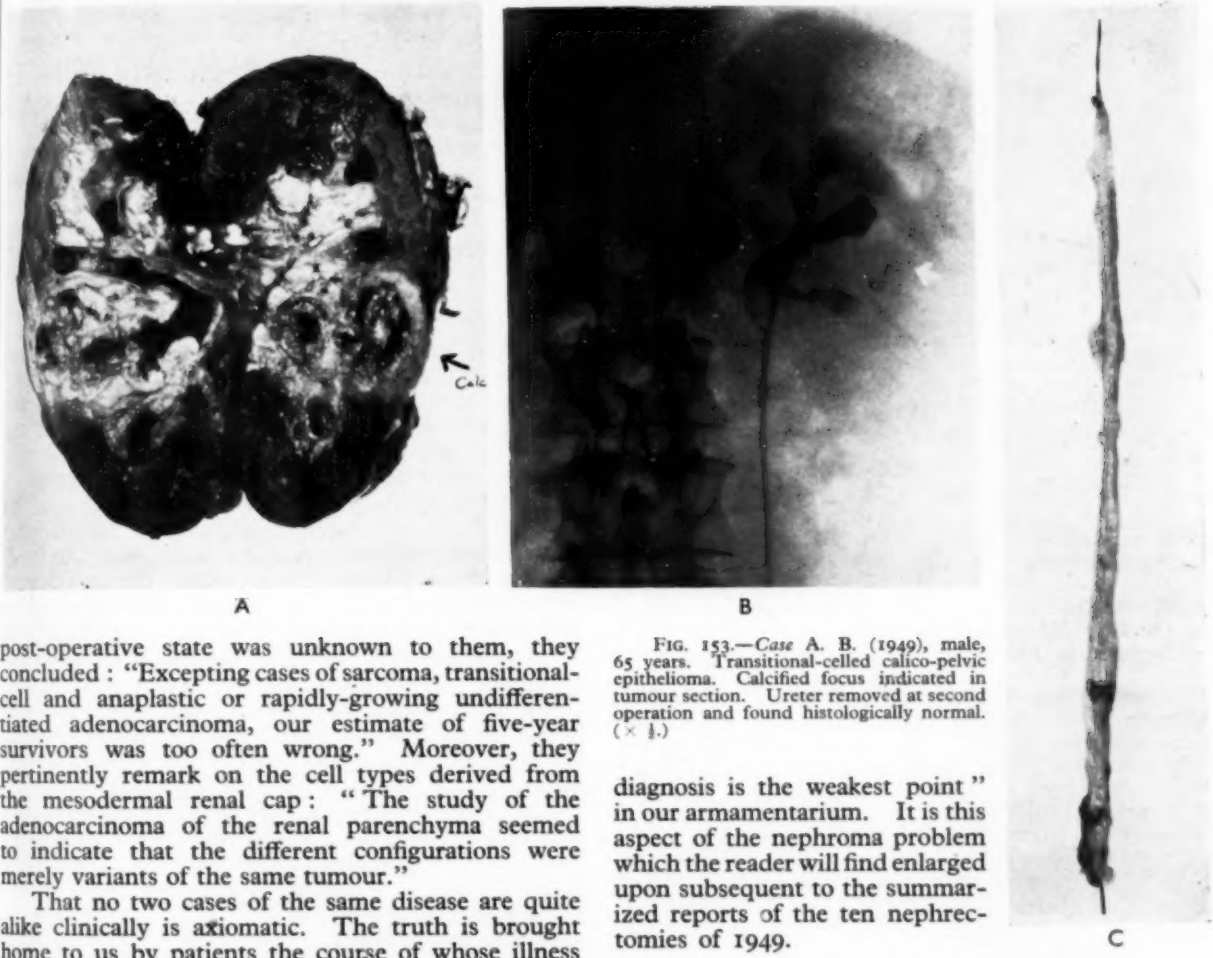
GENERAL REMARKS ON PROGNOSIS

A study of the results of surgery guide us about the patient's prospects after nephrectomy. We judge by the course of illness, findings at operation, and by what the microscope reveals. A rate of 20 per cent of patients surviving five years after nephrectomy was given by me as an average expectation in renal adenocarcinoma. This accords with Barrett and McCague's (1954) report of 20 survivals out of 102 such well-differentiated tumours. The danger of deducing an average figure from too small

a batch of results is usefully exemplified by the surprising 7 five-year survivals out of my own 21 nephrectomies (1949 and pre-1949) for adenocarcinoma. The last quoted authors tested the value histological evidence affords regarding prognosis. Examining tumours of 157 patients whose post-operative state was unknown to them, they concluded: "Excepting cases of sarcoma, transitional-cell and anaplastic or rapidly-growing undifferentiated adenocarcinoma, our estimate of five-year survivors was too often wrong." Moreover, they pertinently remark on the cell types derived from the mesodermal renal cap: "The study of the adenocarcinoma of the renal parenchyma seemed to indicate that the different configurations were merely variants of the same tumour."

That no two cases of the same disease are quite alike clinically is axiomatic. The truth is brought home to us by patients the course of whose illness often shows the widest possible deviation, favourable or otherwise, from our sage predictions. Thus, *Case* W. E. W. in my 1949 group affords a most astonishing and equally pleasant surprise. Existence for a year or two was the most one expected following the operation, yet despite advanced operative findings (*Fig.* 152 A, B) and histology of corresponding virulence, local renal metastases and invasion of renal vein, he was 'himself' and at his usual occupation five years later. Of A. B., the tenth case in my 1949 group (*Fig.* 153), Dr. A. D. Fraser wrote: . . . "rapidly-growing carcinoma of renal pelvis with marked invasion of renal tissue"; nevertheless, the patient, seen in July, 1955, is at work and very well. Such experiences teach the need for warning our patients or their relatives that our forecasts are based on average experience, are only the approximate truth, and may be far from fulfilled in the future of the individual case. But let us, within the bounds of prudence, take care to give the recovering patient the benefit of our uncertainty by indulging his natural hope of a happy issue.

Whether the results of post-operative radiotherapy add significantly to the patient's chances, Barrett and McCague (1954) seem doubtful, on the evidence afforded by their series, but their chief hope, and mine, for improving results lies in timely intervention, and they conclude that "lateness of diagnosis is the weakest point" in our armamentarium. It is this aspect of the nephroma problem which the reader will find enlarged upon subsequent to the summarized reports of the ten nephrectomies of 1949.



A B C

FIG. 153.—*Case* A. B. (1949), male, 65 years. Transitional-celled calico-pelvic epithelioma. Calcified focus indicated in tumour section. Ureter removed at second operation and found histologically normal. (× ¼.)

DIED OF SHOCK

C. H. (F.), 80 years.—This, the sole operation fatality, was the only case of simple tumour.

DIED OF RECURRENCE

W. S. (M.), 67 years.—
History: First hæmaturia *4 years before operation.* Persistent varicocele.
Operation: 1949, clot in renal vein.
Histology: Adenocarcinoma; renal vein invaded.
Sequel: 1953, first illness; metastases obstructed gullet fatally.

A. G. H. (M.), 54 years.—
History: April, 1949, left varicocele treated with suspensory bandage. Recurrent pain left testicle; later, urine became brown; aching lump left loin.
Operation: August, 1949, removal of left nephroma; colon invaded, weight 4 lb.
Histology: Fibrosarcoma; naked-eye appearance of renal, rather than retroperitoneal, origin.
Sequel: March 3, 1950, death from massive local recurrence.

M. A. S. (F.), 64 years.—
History: One week of left loin colic; hæmaturia.
Operation: Aug. 31, 1949, left nephrectomy: pleurotomy incidental. Sept. 29, two pints of sanguineous fluid from left chest—malignant cells absent.
Histology: Adenocarcinoma.
Sequel: Feb. 26, 1952, death from secondaries in spinal column.

R. J. H. (M.), 64 years.—
History: Profuse hæmaturia for 6 weeks.
Operation: Large tumour, local glands, and vena caval deposits removed.
Histology: Adenocarcinoma, clear-cell type; secondary deposits in glands.
Sequel: May 7, 1950, local recurrence; cerebral metastases with hemiplegia; also hæmaturia.

DIED VOID OF RECURRENCE

K. M. (F.), 62 years.—
History: 1949, hæmaturia started. In left nephrogram the kidney shadow, swollen centrally, showed a calcified arc suggesting cyst.
Operation: Left nephrectomy.
Histology: Adenocarcinoma.
Sequel: June 1952, "died in coma following a 'stroke', without evidence of recurrence" (verbal report of Dr. R. M. Ainsworth of Glastonbury).

ALIVE WITHOUT RECURRENCE—5 YEARS AFTER OPERATION

G. T. (M.), 61 years.—
History: 20 months of recurrent hæmaturia (pyelogram normal)—the delay was due to blaming his prostate for the hæmaturia; example of prostatic decoy.
Operation: March 18, 1949, right nephrectomy; surface of kidney suggested caseous tubercles.
Histology: Adenocarcinoma.
Sequel: In latter part of 1954, after herniotomy by Mr. R. V. Cooke, patient complained of backache; radiography showed sclerosing metastases in lumbar spine and markedly in pelvis. Later, coming to me for annual review, he also told a story of prostatism, but was still at work. There was gross evidence of cancerous prostate rectally.
Dr. J. H. Middlemiss (Radiologist) reports that the bony metastases are typical of prostatic, not renal, origin. He started stilbœstrol, since when he seems too busy at work to spare time to see me or to respect the advice of Dr. E. P. K. Rushton, of Turville, about this.

M. A. (M.), 59 years.—
History: Two years recurrent hæmaturia and pain in left loin.
Operation: 1949, transphrenic nephrectomy—pleurotomy.
Histology: Adenocarcinoma.
Sequel: 1955, after pursuit along many devious channels, he was located near Killarney. In a charming letter of appreciation for his well-being he only complained of present inability to find work!

W. E. W. (M.), 52 years.—
History: Somewhat debilitated man with hæmaturia and large loin lump.
Operation: 1949, virulent tumour with scattered satellites in kidney and renal vein.
Histology: Adenocarcinoma, highly malignant (report by Dr. A. D. Fraser).
Sequel: December, 1954, in usual good health.*

A. B. (M.), 65 years.—
History: 1948. Profuse hæmaturia started while he was in hospital with acute tibial osteitis. Clot retention ensued and prostate, deemed responsible, was removed. Fifteen months later profuse hæmaturia with clots; nephroma diagnosed (second example of prostatic decoy in the 7 males of group).
Operation: 1949, nephrectomy. Kidney, centrally, showed a somewhat sunken bossy area, and goblet pelvis.
Histology: Calico-pelvic papilliferous carcinoma. Precautionary ureterectomy; no implantation.
Sequel: Remains at work and found free from recurrence.

In this small series of 10 nephromas, 2 are derived from the transitional-cell lining of the renal pelvis and 8 are parenchymal, about the usual proportion.

One of the 2 pelvic tumours was histologically benign, a title of questionable validity in current clinical opinion. Lund and Lundwall (1956) state: "These tumours are clinically malignant, showing a marked tendency to metastasize and a high incidence of recurrence." In this they are akin to their congeners in the bladder. The other lesion was frank papilliferous cancer. The reported histories show how widely the (parenchymal) adenocarcinoma spreads via the rich venous plexus of the kidney through the general circulation and metastasizes. By notable contrast the outstanding danger with the pelvic nephroma is seeding down the urinary lumen. Happily there were no deposits down the ureter of the patient, A. B. (*Fig.* 153). Yet removal may be respected as a well-merited prophylactic measure for the risk that goes with it is a serious one. Indeed, the whole mucosal surface, bathed by the descending stream of urine, must, for clinical purposes, rank as potentially malignant. Macalpine (1947) stresses this and divides the ureter juxtavesically, using a most scrupulous technique, and fulgurates the vesical segment per urethram later. Higgins (1953) and Lund and Lundwall (1956) are enthusiastic advocates of total nephro-ureterectomy including a cone of bladder. The latter advise this "regardless of the location of the tumour and the histological grade of its malignancy". Like them I find exfoliative cytology of the urinary sediment repays attention in the pelvic lesions while being negative in the parenchymal ones.

The foregoing analytical survey of the 1949 group sheds light upon interesting features about nephroma in adults.

First, the story is a warning to the family doctor on the limitations of bedside diagnosis in bleeding kidney. Among the 10 cases, pain or lump occurred only 4 times.

Secondly, to the surgeon, it is great encouragement to find among the five-year survivors a man of 52 years who was still at full work, whose kidney had a large and, histologically, rapidly-growing carcinoma with satellites and renal vein invasion. Last, but not least, for the urologist, there emerge two matters calling for special and earnest attention. We hope the radiologist, whose science brilliantly illuminates the field of urological diagnosis, will share the following concern.

THE PYELOGRAPHIC FALLACY AND THE PROSTATIC DECOY

There were 7 males in the group and the prostatic decoy caused disaster in 2. In 1 case the surgeon proceeded to do the wrong operation without

* April, 1955: Doctor reports deterioration for 2–3 months, and pulmonary metastases on radiography.

precautionary pyelography of any kind! Moreover the writer has humbly to confess falling into the trap again lately. This episode emphasizes the still more subtle diagnostic fallacy, viz., "pyelogram normal, (gross) nephrocarcinoma present". In my former report, too, was quoted an earlier fatal delay following disarmingly normal pyelography, both intravenous and retrograde. The recent example of this pitfall I want to relate in some detail; it reads rather like a detective story and may quicken urological interest in the weakness of our position as diagnosticians and the resulting delayed extirpation of a deadly tumour.

A. B. (M.), 74 years.—Came in May, 1954, with a well-marked three months' history of prostatism including one bout of hæmaturia just a week prior to the consultation. There was no renal complaint nor swelling—only a plump prostatic adenoma. Urography showed nothing abnormal in the kidneys but gross disease in bladder and prostate (*Fig.* 154). His dysuria increased while he was waiting for admission until August, 1954, when I performed prostato-diverticulectomy. He went home healed about six weeks later with satisfactory use of the bladder. It is relevant that his urine, sampled on Sept. 20, 1954, showed besides pus + + and scanty organisms, *red blood-corpuscles only 1 to 5 per H.P.F.* The infection did not clear as expected, however, and on Dec. 20, he came for review, done by my registrar, Mr. Keith Chitty, who deferred cystoscopy pending toilet of infected phimosis and some meatal stenosis. The next news of him was in March, 1955. There was hæmaturia culminating in clot retention: a transient attack in mid-December was unreported to me. Thus bleeding had been absent for seven months since the initial bout of hæmaturia.

March 29, 1955: Seeing him the morning after admission he looked well enough, though possibly there was a little loss of weight and usual vitality. The belly felt normal. The lower pole of the right kidney was, perhaps, palpable, The penile condition was as before. Urine normal, naked eye; he said function was all right apart from the hæmaturia. Rectally a vestigial capsule was the only legacy of the former prostate. Excretory pyelograms, taken elsewhere (Feb. 28, 1955), showed the same pattern as pre-operatively (*Fig.* 155—May, 1954) except for an ominous abrupt truncation of the right inferior calix (*Fig.* 156). *March* 30: First chromocystoscopy and segregation of urines. No source of hæmaturia visible in the lower tract, but a thin dribble of rich red blood from left ureter. Catheter from right kidney dripped clear blue urine early. On the left 5 c.c. of colourless urine was aspirated, becoming very faintly tinged red. Two days later, on cystoscopy, instead of a trail of blood, a horn-shaped clot was seen immovably adherent by its tip in the mouth of the left ureter. Meantime the pathologist reported only scanty R.B.C.s and W.B.C.s in right ureteric urine, and R.B.C. + + + with scanty W.B.C.s in left ureteric urine. The *left* kidney now seemed the guilty one, reversing the natural inference from the February pyelograms which showed failed filling of lower calix of *right* kidney! Contradictory findings and a fervent desire for sharp delineation of renal patterns led to retrograde pyelography; pictures were normal (*Fig.* 157) apart from mottling of left kidney pelvis and adjacent part of the upper calix, possibly indicative of blood-clot. However, a patient suffering recurrent clot retention and at two cystoscopies bleeding only from the left ureter called for renal exploration.

FIG. 154.—*Case* A. B. Cystogram.

FIG. 155.—*Case* A. B., May, 1954. I.V. Pyelograms classed as normal.

FIG. 156.—*Case* A. B., February, 1955. I.V. Pyelograms—R. lower calix absent.

April 1, 1955: Left nephrectomy for obvious Grawitz tumour at upper pole and nodules dispersed throughout the kidney (*Fig.* 158). Histology, adenocarcinoma. He now has easy bloodless urination.

The foregoing tells of a lurking nephroma missed by two urologists, my colleague Mr. Ashton Miller, and myself—and of an associated adenomatous

prostate being wrongly blamed for hæmaturia. Blinded by this, I had proceeded to prostatodiverticulectomy. True there was collateral dysuria meriting operation, but at the proper time. Instead, seven months elapsed before the patient returned to me with the recurrent hæmaturia, and even then the responsible renal culprit evaded pyelographic detection. Let us ask ourselves why such mistakes happen.

FIG. 157.—*Case* A. B., March, 1955. Retrograde pyelograms. Caliceal patterns normal.

Is not the answer that we have become slaves to scientific apparatus, acting on its findings without due discrimination? For, although we are deeply indebted to radiology and its increasing diagnostic range, and impressed by the fact that "the camera cannot lie", we must not forget to use the "pinch of salt". The unpleasant fact is we often intervene late, which ill becomes a profession pressing for *early diagnosis in cancer*.

Let us label renal cancer a dark horse at the outset and plan our clinical approach accordingly. To the *student* a sudden bout of hæmaturia in the adult should signalize potential nephroma; he should view it like a fireman does a fire alarm! Among the 10 nephromata of the 1949 group, 9 were cancers; all had heavy hæmaturia at the start—the typical, though not invariable, onset. A localizing clue, like pain or lump, occurs in the minority; therefore, in practice we need to be on the *qui vive* either with the symptomless attack out of a blue sky or when a bout of bleeding supervenes during an illness. To the *patient*, sinister implications excusably escape him when bleeding is symptomless and deceptively intermittent; he drifts. To the *doctor* its possible origin in relatively rare renal tumour may be forgotten; or, if it happens during illness, he is apt to regard it as a complication.

MODERN UROLOGY AND THE BOUT OF HÆMATURIA

Nephroma is uncommon, but during the years ahead of him a few dozen cases will pass through the hands of a young urologist. He can catch them early, instead of regrettably late, if he learns from his predecessors' mistakes. Moreover, his timid patients may be helped, too, by telling them that E. D., a man of 48 years, had nephrectomy in 1939 as did L. H., a woman of 44 years in 1934—both for adenocarcinoma—and that they are alive and well in 1955. (Kidneys figure in the 1949 report.)

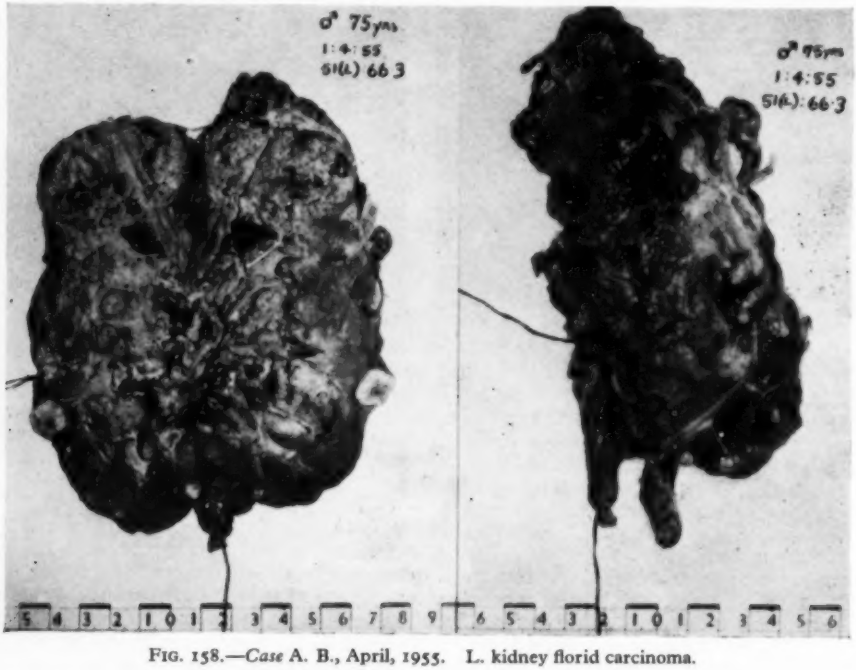


FIG. 158.—*Case* A. B., April, 1955. L. kidney florid carcinoma.

Nephroma's Bloody Début.—The sudden bout of hæmaturia is a challenge to our diagnosis. When the doctor is confronted with it and no cause is apparent, he should telephone the urologist " cystoscopy urgently wanted " ; or, if illness is already present, whoever is already treating the patient should be informed and regard it as an alarm signal calling for prompt open-minded examination of his patient.

What is to be the urologist's attitude ? " Take no chances ". Clinical examination inconclusive, it is his bounden duty to do urgent cystoscopy for *to spot a bloody efflux from a single ureter* is the most influential contribution to the timely diagnosis of nephroma. It is the key to the situation. The evidence is fleeting and, if missed, may not recur for months. When this golden opportunity offers and he takes advantage of it, the urologist's conscience will be clear. Too late for this, he does the next best thing and moves to the routine ritual of full urological investigation ; but he must not then ' go to sleep ', for some indispensable responsibilities remain which he cannot off-load on to his willing colleagues in pathology, radiology, etc. Examination under anæsthesia and chromocystoscopy are usefully included and also retrograde pyelography. The status of radiography with perirenal insufflation and aortography in this connexion has yet to be assessed. *Where findings are negative*, the next step may well be decisive in the fate of nephroma.

Hæmaturia is dramatic and may seem to demand surgery, if no other treatment is appropriate. But though it may be hard to refrain, the urologist must be wary of doing something merely because it is hopeful and, anyway, may be generally helpful, e.g., speculative prostatectomy. Three instances have been given of a prostatic decoy since 1949. He well may be incurring professional disrepute by his intervention ; also, symptoms returning, it clouds the issue and hinders subsequent amendment. Instead, what is called for *when diagnosis is unsettled* is reflection and cogitation.

Obviously, ureteric bleeding of obscure bilateral origin, or in, say, a hæmophiliac is off the surgeon's beat ; but *per contra* caution is needed in treating expectantly a patient diagnosed as having renal epistaxis, a very different category from its nasal counterpart. At the end of a thorough investigation, medical science may claim to have excluded the surgical causes of a bout of hæmaturia ; but the naked truth, as exposed by my foregoing stories of relevant cases, is that the ominous possibility of lurking nephroma with which we began the search remains and is undiminished by the proved absence of alternative causes.

Faced with the frank dilemma due to fallacies of pyelography, etc., two courses are open :—

1. Expectant treatment, viz., three-monthly review, and warning the patient that immediate cystoscopy is vital when bleeding starts again.

2. Diagnostic surgical exposure of the kidney(s). This innovation, regrettable as it may sound, seems an inevitably logical conclusion to have in mind for a certain number of cases.

The urologist has a grave responsibility in deciding which of the above two lines of action to adopt and must bring judgement and experience to bear on each individual case. Where he courageously advocates surgery, he will expose the more suspicious side ; and, without this lead, the usually more accessible right. The kidney, though looking externally normal, may feel thickened centrally, calling for biopsy. Otherwise the peritoneum affords access for palpation of the opposite kidney, and both ureters for possible tumour. This locates or excludes the suspected neoplasm of the urinary tract and leads to the necessary eradication or simply to closure. In the latter case he reassures the patient and eases the mind of both parties after trauma involving slight risk, whereas in the former his logical act has discovered a lethal lesion and enabled him to do a timely nephrectomy instead of the belated operation which too often happens with present methods. Where pathology, definite or dubious, is felt in the opposite kidney it may be best, because of the general condition of the patient, to deal with this on a separate occasion after the exploratory incision has healed.

SUMMARY

1. Experience proves that it needs a first-class surgical detective to discover the deceptive cancer of the hypochondriac recess early in its history. Too many of the ablations of nephroma are avoidably belated.

2. Let us spread abroad the crucial importance of immediate cystoscopy when an adult is taken with a sudden bout of unexplained hæmaturia.

3. The disquieting delay in operating on nephroma results from a too implicit trust in data derived from routine urological investigation. *Negative findings are not infallible !* Despite them, the risk of tumours remains in a significant proportion.

4. Diagnostic exposure of the kidney will settle the diagnosis and should be done early where grave suspicion of lurking nephroma remains. The risk attendant on surgical incision is in these days no greater than that incurred by expectant treatment.

5. A plea is made for the urologist to open the loin on occasion with no more ado than attends the familiar abdominal exploration by the general surgeon.

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VOLVULUS IN WESTERN INDIA

A CLINICAL STUDY OF 40 CASES, WITH PARTICULAR REFERENCE TO THE CONSERVATIVE TREATMENT OF PELVIC COLON VOLVULUS

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INTRODUCTION

Commenting on a series of cases of small-intestine volvulus reported from Kenya by Kerr and Kirkaldy Willis (1946) Sir Heneage Ogilvie writes: "South African anatomists have told me that (in the Africans) anomalies of intestinal rotation are more common, particularly the persistence of unfixed mesenteries. It is possible that it is in the greater mobility of the alimentary canal rather than in his dietary habits that the reason for the proneness of the African to volvulus is to be found."

Experience in the western part of India for fifteen years indicates that volvulus, both of the small and large intestine, is common here also, and that probably both congenital abnormalities and diet play a part in its causation.

The relative importance of these factors in aetiology, suggested by a series of 40 cases, is considered and a clinical account of diagnosis and treatment given.

Geographical and Local Conditions.—The region in which these cases were observed lies on the western border of the central India plateau of the Deccan, about 200 miles inland from Bombay.

The patients were seen in a 100-bed mission hospital maintained by The Salvation Army in Ahmednager city, which is the administrative centre of an agricultural district with a population of about 1,400,000. The district extends about 35 miles in one direction and up to 100 in others. Sixty to seventy per cent of the patients are villagers from this extensive area, the remainder being from the city (80,000) and vicinity.

The surgical work of the district is shared with the Government District Hospital (80 beds) and two smaller private hospitals. However, there is a strong impression (reliable statistics being non-existent) that only a fraction of the village people requiring surgical attention attend hospitals for treatment. The urgency of the symptoms of acute intestinal obstruction probably accounts for a relatively high incidence of this emergency in the work of the hospital.

INCIDENCE

1. Intestinal Obstruction.—During the years 1948–53, 168 cases of acute or subacute intestinal obstruction were treated, this being the commonest abdominal emergency at the Hospital (appendicitis 66, ectopic gestation 17, perforated duodenal ulcer 3).

The figure of 168 represents about 2½ per cent of surgical admissions, compared with 0·85 per cent of admissions for intestinal obstructions to the surgical wards of the Massachusetts General Hospital (quoted by Wangensteen, 1949). It is noteworthy that the actual number of admissions for intestinal obstructions at our much smaller hospital is practically the same as that of the Massachusetts General Hospital (168 in 5 years, E.B.H.: 335 in 10 years, M.G.H.).

2. Volvulus of All Types.—Volvulus was the cause of obstruction in 40 of the 168 cases of intestinal obstruction, that is 24 per cent.

This high figure agrees with other series in India—Ojha (1950) 19 per cent; Bhargava (1947) 37 per cent; McWatters (1945) "20 per cent of small-intestine obstructions excluding strangulated hernia". It contrasts strongly with the incidence in reported large British series—Souttar (1925) 2·4 per cent; Vick (1932) 2·5 per cent; and in American series—Wangensteen (1949) 4 per cent; Henry (1940) 7 per cent in non-malignant obstructions.

On the other hand, a still higher proportion is reported from Russia and Eastern European countries—Obalinski (1894) 35 per cent; Perlman (1925) 52 per cent (both quoted by Wangensteen, 1949), while Braimbridge (1946) is reported by Kerr and Kirkaldy Willis (1946) to have stated that "in 25 years of surgical practice in Kenya he has found it the commonest cause of intestinal obstruction in the native, occurring in more than half the cases".

The incidence in India therefore appears to be between the much lower incidence in Britain and the relatively low incidence in the U.S.A., and the still higher incidence in Russia, Eastern Europe, and Kenya.

3. Varieties of Volvulus.—The table shows a marked difference in the incidence in different countries of the three varieties of volvulus considered. Rarer varieties, such as in the stomach, transverse colon, and volvulus neonatorum, have not been

Volvulus in Different Countries

Country	Series	Number in Series	Site		
			Cæcal (Right Colon)	Small Intestine	Pelvic Colon (Left Colon)
			per cent	per cent	per cent
Great Britain	Souttar (1925)	77	23	50	27
„ „	Vick (1932)	176	20	48	32
U.S.A.	Sweet (quoted by Wangensteen 1949)	52	12	68	20
Scandinavia	Bruusgaard (1947) Frimann Dahl (collected series) (1954)		10–20	25–35	60–65
India	Andersen (1956)	40	5	35	60

encountered in the series presented, and are not further considered.

It is very suggestive that Moretz and Morton (1950) writing from Rochester, N.Y., reporting a series of 36 cases of small-intestine volvulus, found that 25 of them had undergone previous abdominal operations and that in 23 the volvulus was considered to be due to post-operative adhesions. In the present series only 3 out of 14 attacks of small intestine volvulus were due to post-traumatic adhesions (2 post-operative, 1 following bullock gore), the rest occurring " naturally ".

In comparing the percentages of small-intestine volvulus, therefore, the high number of post-operative cases, especially in the U.S. series, must be taken into account. Even after allowing for this, however, the proportion of pelvic colon volvulus in India and Eastern Europe and Russia appears to be relatively high and suggests a common factor not found in Britain and the United States. A bulky, high-residue vegetarian diet may be an important aetiological factor.

Sex.—It is surprising that of 33 patients only 4 were women and they were all in the pelvic colon group. This suggests another factor or factors and agrees with the general experience. Bhargava (1947) notes a proportion of 3 males to 1 female. A possible explanation is the laxer abdominal wall especially after pregnancy and the wider female pelves, and possibly, in this series, as suggested by Bhargava " reluctance (due to social causes) on the part of the old women and their relatives to seek institutional treatment ".

AETIOLOGY

Three major factors therefore present themselves for consideration as predisposing causes—namely, adhesions due to previous inflammations, congenital anomalies, and diet.

1. **Inflammatory Adhesions.**—In 3 cases there were definitely inflammatory bands to the parietal peritoneum following: (*a*) operation for retained testis; (*b*) bullock gore; (*c*) unoperated appendicitis. The mechanism of these parietal bands in causing volvulus is well recognized (Rodney Smith, 1948).

2. **Congenital Anomalies.**—The importance of congenital abnormalities varies with the part of the bowel concerned.

a. Cæcum.—In the case of the cæcum this is a *sine qua non*. Anatomical studies by Harvey (1918) showed abnormalities in the cæcal attachment from defective completion of the third stage of rotation (Dott, 1923) in 13·3 per cent of 105 infants. Wolfer, Lindsay, and Barry (1942) found 11·2 per cent with a freely mobile cæcum and " prefixation of the terminal ileum " in 10·4 per cent of a series of 125 adult cadavers.

The usual type of abnormality is well illustrated by the single patient in the series, who had a mobile cæcum and ascending colon with a common ileo-cæco-colic mesentery. In addition a definite long peritoneal band extended from the right iliac fossa to the posterior aspect of the mesentery near the bowel attachment about 2 in. above the ileocæcal valve (*Fig.* 159).

b. Small Intestine.—In 6 cases out of 14 there were evident congenital abnormalities. In 4 there was a band which passed from the terminal ileum or the under surface of the mesentery adjoining, 1–5 in. from the ileocæcal valve, to the right iliac fossa. In none of them was there evidence of previous appendicitis or other inflammation. They were associated with a mobile cæcum which was drawn markedly to the midline by the volvulus. A mobile cæcum without any band was noted in 2 more cases (*Figs.* 160–163).

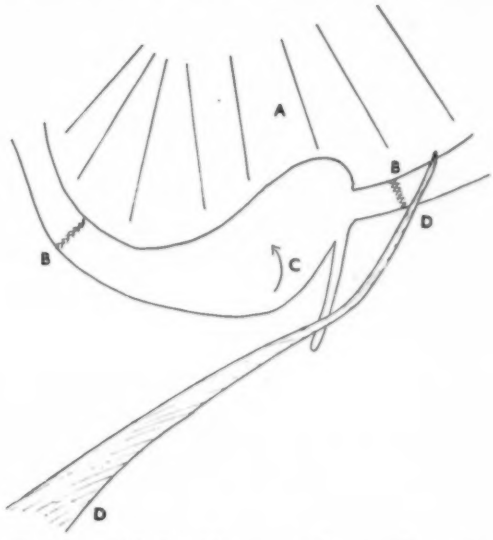


FIG. 159.—*Volvulus of Cæcum.* A, Common ileocæco-colic mesentery; B, Congestion at sites of torsion; C, Cæcum and terminal ileum lifted upwards to show: D, Firm peritoneal band from posterior aspect of a mesentery about 2 in. above ileocæcal valve to the right iliac fossa.

Such a mobile cæcum, whether associated with a band or not, because the cæcum is less firmly fixed than normal to the posterior abdominal wall, implies conversely that the terminal ileum is more fixed than normal by the lower attachment of a short mesentery, corresponding probably to the " prefixation of the terminal ileum " referred to by Wolfer, Lindsay, and Barry (1942). Such a relatively fixed point would predispose to a volvulus compared with the normal attachment of the terminal ileum to the medial baggy wall of the cæcum, which would allow some play.

This suggestion of the significance of precæcal ileal fixation is supported by the fact that in the series of 168 cases of intestinal obstruction of which this is a part, there were 4 other similar precæcal bands which caused simple obstruction at this site, and in one case local narrowing due to fibrosis extending into the mesentery, probably following previous attacks relieved spontaneously.

Further, in the series there were 7 other cases of bands, apparently congenital, with no evidence of past or present inflammation, causing obstruction in other parts of the intestinal tract.

These findings suggest a relatively high proportion of congenital peritoneal abnormalities in India (although the large " pool " from which these cases are drawn must be kept in mind).

The precæcal fixation point was observed to be the exact site of the lower limit of the twisted bowel

in these cases, while the upper limit varied from 2½ to 10 or 11 ft. above this. All the cases described had a definite pedicle in the precæcal region and other cases of some degree of bowel rotation due to bands are not included.

Other series have also commented on the importance of "fibrous bands, presumably congenital" in the aetiology of small-intestine volvulus. Moretz and Morton (1950) reported 7 out of 13 cases, excluding the post-operative adhesions. Ojha (1950) noted 3 bands in the ileocæcal region out of 13 cases, not including those due to tuberculosis. Tanner (1946) in 13 cases at the London Hospital in 10 years found 7 bands, 3 being from a Meckel's diverticulum. Practically no information regarding the average length of the small intestine and height of the mesentery in India has been found, but routine post-mortem examinations to provide some information are being conducted at the Vellore Christian Medical College, Anatomy Department (Professor L. Chacko, 1955). A few preliminary examinations show no marked deviation from normal.

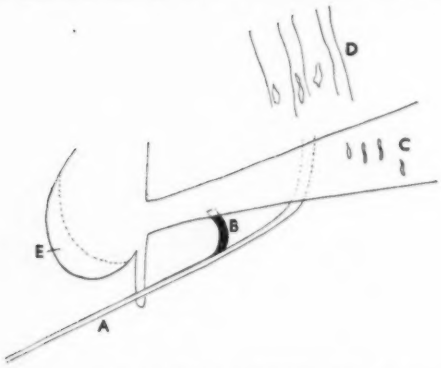


FIG. 160.—*Volvulus of Small Intestine* (Band). A, Tense band along brim of pelvis merging with inferior leaf of mesentery; B, Subsidiary band extending into posterior ileal surface; C, Submucous hæmorrhages. D, Glands in congested mesentery; E, Partly mobile cæcum.

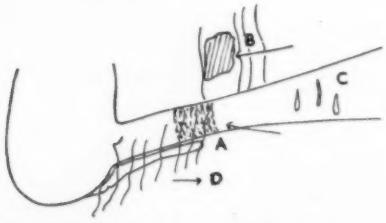


FIG. 162.—*Volvulus of Small Intestine* (Post-appendicitis adhesions). A, Congestion at torsion site; B, Superficial ulceration of mesentery; C, Submucous hæmorrhages; D, Post-inflammatory adhesions.

c. Pelvic Colon.—The important predisposing cause in volvulus of the pelvic colon is a long sigmoid loop. There is a natural variation of this within a wide range (Cunningham (1937) 5–35 in., average 16 in.).

An interesting paper by Aiyar and Bobji (1943) points out that in Madras, on the east coast, the average length of the pelvic colon measured in 30 unselected dissection cadavers (formalin preserved) was 9 in. only, and the height of the pelvic mesocolon was 2½ in. There was only one long pelvic colon, measuring 19 in.; all the rest were between 4 in. and 13 in. This rather surprising finding is further borne out by Chacko (personal communication), where in a few unselected cases the average pelvic colon was 13 in.

However, much more extensive examinations will be necessary before conclusions can be drawn, particularly one series in different areas of the length of the pelvic colon in a significantly large number of children to determine congenital differences, and another series in the same areas of the length after 35 years to determine acquired lengthening. Previous studies have indicated that there is possibly a racial or a regional factor in that a higher proportion of people in some countries have a long sigmoid loop. For example, Samson (Russia) found 20 per cent of long pelvic loops, compared with only 4 per cent by Curschmann in Germany (quoted by Wangensteen, 1949). These figures are in relation to the incidence of volvulus in these two countries. However, it is not clear whether these differences are congenital (racial) or acquired (regional, probably dietary).

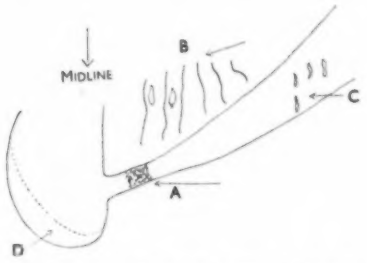


FIG. 161.—*Volvulus of Small Intestine* (Mobile cæcum). A, Congestion at pressure site; B, Thickened mesentery and enlarged glands; C, Submucous hæmorrhages; D, Partly mobile cæcum.

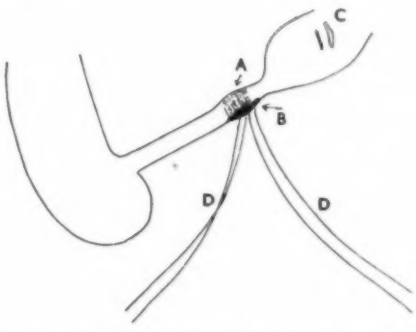


FIG. 163.—*Volvulus of Small Intestine* (Local necrosis at pressure site. Bands). A, Congestion; B, Linear pressure necrosis; C, Dilated ileum; D, Bands.

Another congenital factor is the *attachment of the pelvic mesocolon*, an open (wide) attachment reducing the likelihood and a closed (narrow) attachment increasing it by bringing the ends of the loop closer together.

3. The Influence of Diet.—Several observations suggest the importance of this. Of the 33 patients, 30 were farmers or farm labourers (90 per cent). The average proportion of these in the hospital is about 60 per cent. No Mohammedans (normally 20 per cent of the population, and meat eaters) or middle-class town Hindus such as merchants (whose diet is more varied) were included.

The villager eats up to 2 lb. of cereal a day, usually millets called bajri or jowar, with a little green vegetable, and some chillies. At a certain time of the year he eats the fresh grain slightly roasted, and an attack has been noted to follow some hours after this in one or two cases. Otherwise the grain is ground and he prepares thick unleavened bhaker (flat baked cakes). His bowel habits are very regular, but it is certain that his pelvic colon reservoir would contain a bulkier residue than that of a habits from a mixed to a mainly coarse vegetarian one (Bruusgaard, 1947). Frimann Dahl (1954), however, points out that it has remained high, although somewhat less, in the post-war years, so he does not consider the relationship with the changed diet conclusive.

It is at least, however, suggestive, and the war-time diet effects may be protracted.

It is also noted that the average age of the pelvic colon group in this series is 50, while that of the small

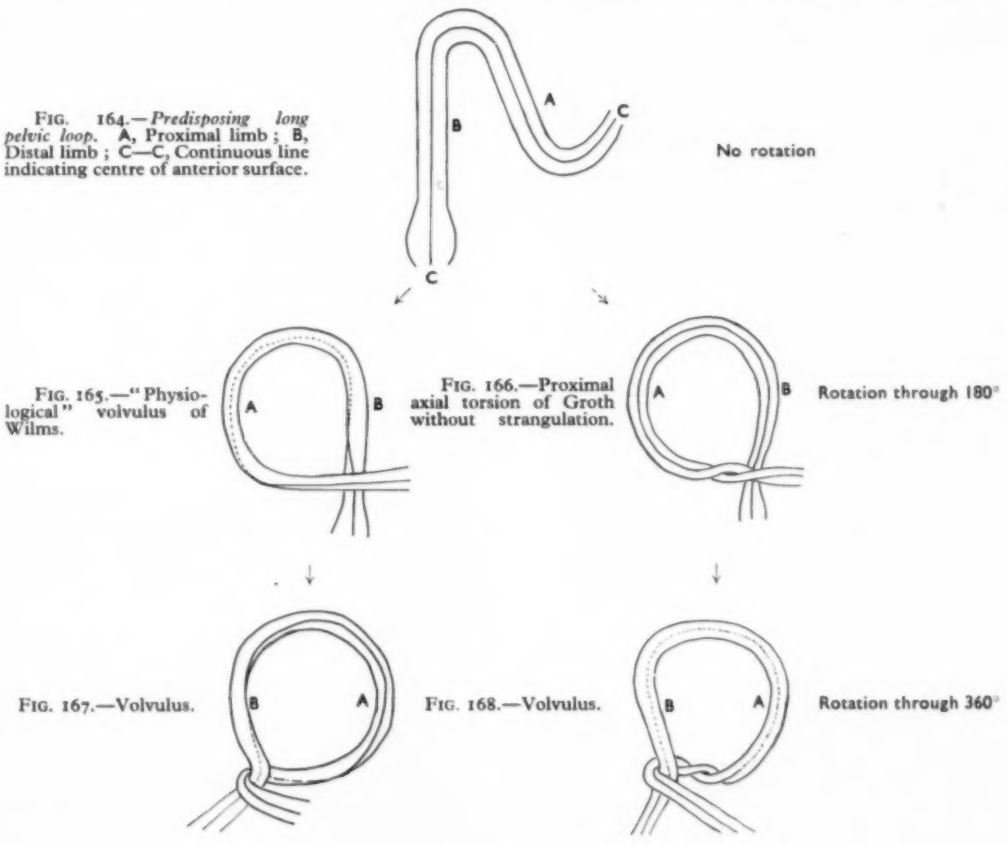


FIG. 164.—*Predisposing long pelvic loop.* A, Proximal limb; B, Distal limb; C—C, Continuous line indicating centre of anterior surface.

FIG. 165.—"Physiological" volvulus of Wilms.

FIG. 166.—Proximal axial torsion of Groth without strangulation.

FIG. 167.—Volvulus.

FIG. 168.—Volvulus.

FIGS. 164–168.—*Mechanics of Volvulus.*

non-vegetarian. Kirk (1937), dealing with the disposal of sewage under tropical conditions, states that an average allowance of 16 oz. daily must be made for vegetarians compared with 6 oz. daily for non-vegetarians. This loading of the pelvic colon reservoir might cause an elongation of the pelvic colon and mesocolon associated with narrowing of the mesentery of the posterior wall (Aird, 1950), and also the ends of the pelvic loop might be brought closer together.

It is of interest that, when comparing experiences with two Indian colleagues practising surgery in Sholapur, 300 miles farther south, one, with a comparable class of village patients, had a similar large number of cases of volvulus, while the other, with a private clinic mainly for townspeople, hardly saw any.

In Norway a special circumstance supports this view of the acquired predisposition due to diet. The incidence rose steeply during the German occupation when there was a compulsory change of dietary intestine group is 39, suggesting more long-acting acquired causes in the colon group.

In Europe the high incidence in mental asylums may be related to overeating and the eating of indigestible substances.

PATHOLOGY

Secondary Pathological Changes.—No recurrences or secondary changes due to previous attacks were observed in the small-intestine cases, although they are described by McWatters (1945).

They were, however, marked in the pelvic colon group, and the following changes were noted:—

a. Thickening and sometimes narrowing of the pelvic mesocolon.

b. Gross fibrosis evidenced by macroscopical criss-crossing white fibres (in several cases).

c. Stenosis of the ends of the bowel (very marked in 1 case), and dilatation and hypertrophy of the rest

of the loop—the so-called ' omega shape ' (Frimann Dahl).

d. Secondary attachment of the apex to the parietal peritoneum (1 case).

These changes are the result of previous attacks, the majority spontaneously relieved, and some surgically. They indicate a strong tendency to further recurrences. (Present series, 24 attacks in 18 patients; Bruusgaard (1947), 168 attacks in 91 patients.) It also suggests an explanation of the relatively benign nature of recurrent attacks, the blood vessels being mechanically protected by the thickened mesentery, so that the onset of gangrene is late. In fact, probably the most dangerous attack is the first one, and in such cases, gangrene can occur very early (*see Fig.* 172).

Mechanics of the Pelvic Colon Volvulus (*Figs.* 164–168).—Wilms (1906) has pointed out that with a long pelvic loop, a physiological volvulus of 180° may occur without symptoms, and is the starting point of the clinical attack. (*Fig.* 165.)

Groth (1934) has further pointed out that such a twisting (180°) of the sigmoid and mesocolon is associated with an axial torsion of the bowel wall of twice the amount (360°) (*Fig.* 166). This can be readily demonstrated with a bicycle tube. If this axial torsion is spread evenly over the length of the bowel, then there is no obstruction: but if the torsion is at the ends, or especially at one end only, then mechanical obstruction arises and causes symptoms. However, this condition is fairly easily reversible in the early stage and may account for mild attacks which are spontaneously reduced. If, however, due to subsequent hyperperistalsis and a loaded colon a further twist of 180° in the same direction occurs, then it rapidly progresses to a condition which is reducible with difficulty, or not at all. When distension is extreme, a condition arises which has been aptly described as " a balloon tyre clamped between the anterior and the posterior abdominal walls " and the condition becomes irreversible without external aid.

Frimann Dahl gives the approximate percentages of the different degrees of rotation as 180°, 35 per cent; 360°, 50 per cent; and 540°, 10 per cent.

The direction of the rotation as observed in the series has been variable, the clockwise direction being more common (*Figs.* 164–168).

The suggestions regarding the relative influence of congenital abnormalities and diet, and secondary changes tending to recurrence in the three groups can therefore be summarized as follows :—

	Cæcum (Right colon)	*Small Intestine*	*Pelvic Colon (Left colon)*
Congenital factors	+++	++	+
Dietary factors	+	+	++
Secondary changes tending to recurrence	++	+	+++

GENERAL CLINICAL PICTURE

During the period of 5 years in which these cases were seen, certain broad clinical pictures were gradually appreciated.

Excluding for the moment cases with gangrene, there was an evident distinction between the pelvic colon and the small-intestine group.

In the pelvic colon group the distension was maximal, but the general condition surprisingly good; while in the small-intestine group the distension was moderate but the general condition much poorer. This is no doubt related to the greater degree of dehydration and disturbance in the electrolyte balance in the small-intestine group and possibly greater absorption of the toxic peritoneal exudate (Aird, 1941), which also characterizes this group.

Cases with massive gangrene, whether of the pelvic colon or the small intestine, presented a much more severe picture of grave surgical shock.

Regular use was made of plain radiographs immediately after admission, which were of the greatest use in supplementing the clinical diagnosis.

CLINICAL AND RADIOLOGICAL DIAGNOSIS AND TREATMENT

PELVIC COLON—VOLVULUS

The pelvic colon group will be considered first as it is the largest and its clinical picture was appreciated first. A distinction will again be made at once between the usual more benign type and the type with early gross gangrene.

A. ' BENIGN ' TYPE

Clinical Picture.—The usual *history* given was of sudden severe abdominal pain associated with vomiting 2–5 days before admission. The patient also complained of constipation, inability to pass flatus, and rapidly increasing distension. Vomiting usually had ceased after the first day and the abdominal pain on admission was more or less constant, generalized, and fairly severe (associated with the torsion of the mesentery and the extreme distension); there was often a history of previous attacks of similar pain ending spontaneously, and one patient (who proved to have no urinary obstruction) complained chiefly of attacks of retention of urine.

On *inspection* the patient had the most extreme type of distension, which was characteristically more marked in the upper than in the lower abdomen, and there, more on the left than on the right side. Sometimes it was possible to see clearly the contour of the large double pelvic loop passing up to and disappearing under the left costal margin.

The pulse volume and rate (80–90) and general condition of the patient were surprisingly good, contrasting with the extreme distension. *Palpation* confirmed the findings of inspection, although sometimes (when the remainder of the colon or small bowel was much distended) no definite loop was palpable. On one occasion gross peristalsis was felt and seen, but this was not usual. Percussion gave variable findings and free fluid was not marked.

Auscultation gave occasional sounds of hyperperistalsis, but not the well-marked periodic sounds of a simple obstruction without strangulation, volvulus, or secondary paresis.

Radiological Diagnosis.—A plain radiograph in the standing position with horizontal rays was taken as a routine (without an enema beforehand)

and gave a characteristic picture which led—with the clinical picture—to a correct diagnosis in all save 2 cases. In one, the absence of the usual fluid levels caused doubt, and in the other, a volvulus was diagnosed when there was an abnormal opening in the base of the small-intestine mesentery through which the pelvic colon passed and was strangulated, which simulated volvulus closely.

The radiograph findings (*Figs.* 169–172) observed were :—

1. A grossly distended and easily seen double pelvic loop with differential fluid levels, giving rise to the appearance described aptly as a ' pair of scales '. In the cases of gangrene, the wall outline was unusually well defined (*Fig.* 172). It is important, especially when gangrene is suspected, to examine for free gas under the diaphragm due to perforation.

FIG. 169.—After reduction of a case conservatively, showing elongated and dilated pelvic loop with narrowed ends, the so-called Ω (omega) shape.

FIG. 170.—Plain radiograph. Circular pelvic colon loop (less typical) in a woman.

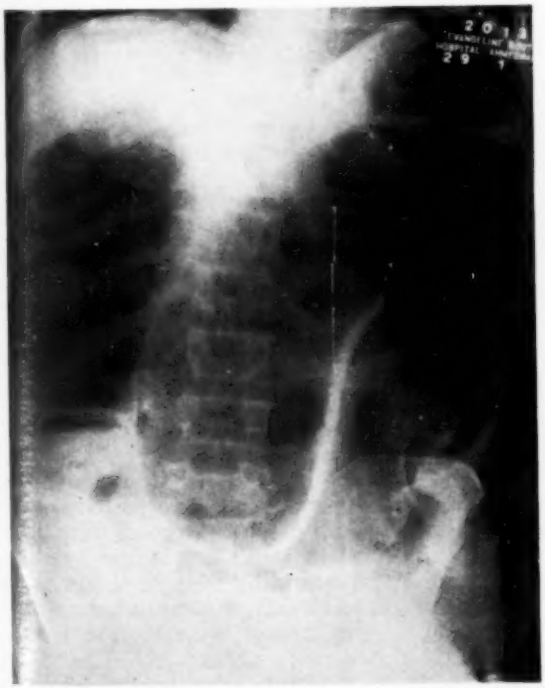


FIG. 171.—Plain radiograph. Pelvic loop of usual contour, but without fluid levels. Also gross dilatation of rest of the colon, especially cæcum.

FIG. 172.—Plain radiograph. Advanced gangrene of pelvic loop. Note gross distension, well-defined wall, and marked thick irregular line between the two limbs.

In this case a barium enema is contra-indicated (Frimann Dahl).

2. A variable amount of gas and fluid in the remainder of the colon, and in 1 case gross distension of the cæcum (*Fig.* 171).

3. Occasionally a few fluid levels in the lower small-intestine loops.

The appearance is so characteristic that diagnosis is not usually in doubt. However, a cæcal volvulus

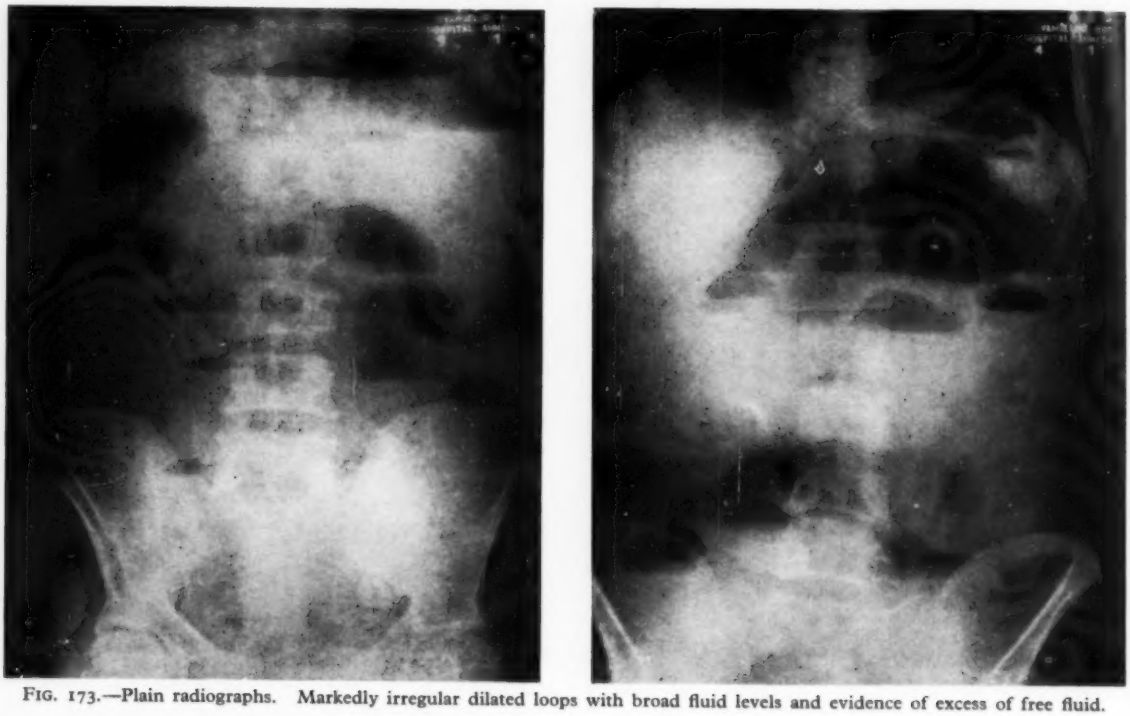


FIG. 173.—Plain radiographs. Markedly irregular dilated loops with broad fluid levels and evidence of excess of free fluid.

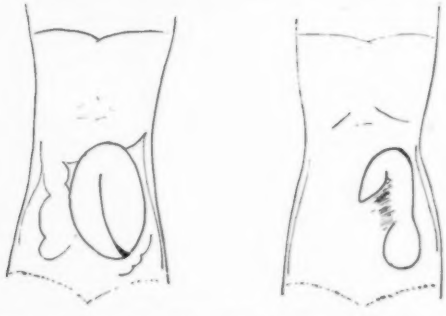
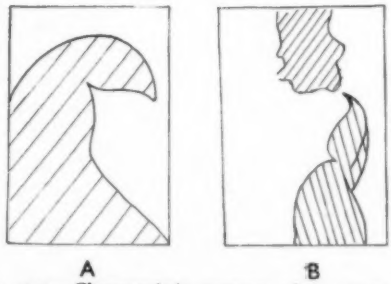


FIG. 174.—Representation showing difference between sigmoid and cæcum volvulus (*after Frimann Dahl*).

may be displaced over to the left and simulate the condition closely with two fluid levels (Frimann Dahl, 1954), but here the loop is 'open' towards the right, while the pelvic loop is 'closed' and points to the left iliac fossa (*Fig.* 174). Occasionally very distended small-intestine coils may give rise to uncertainty as to whether or not there is colon obstruction present. Here a barium enema is given, the catheter not being introduced beyond the rectum. The appearance at the pelvirectal junction at the root of the volvulus is typically that of "the bill of a bird of prey", but rosette or other formations are described (Frimann Dahl (*Fig.* 175 A)). Also, when the twist is loose, there may be a 'spill' above it (*Fig.* 175 B). Observation of the direction of the 'beak' is of value in the conservative method of treatment. Further, a lateral barium radiograph of the rectum will determine accurately the distance from the anus to the twist.

Treatment.—This requires consideration under two headings, namely: (1) emergency treatment of the attack; (2) curative treatment.



A B

FIG. 175.—Characteristic contour of the rectum at the site of torsion (barium enema). A, Without spill (usual); B, With spill.

1. *Emergency Treatment.*—While the usually accepted treatment is by laparotomy and reduction of the pelvic volvulus under vision after decompression by a flatus tube from below, experience has given grounds for increasing confidence in Schilling's conservative method of reduction in the 'benign' type as defined here, *especially when there is a history of previous attacks either relieved spontaneously or by treatment.*

Technique of conservative reduction: The patient is placed without any anæsthesia in the lithotomy position with the buttocks well over the edge. A standard sigmoidoscope (25 cm.) is passed carefully

with adequate illumination (and swabbing if necessary) till the site of the twist is reached, usually between 15 and 25 cm. from the anus.

With the sigmoidoscope opening just below and pointing in the direction of the twist, a flatus tube 1·0 cm. wide, 50 cm. long is passed gently through the twist. The result is a not unwelcome bath of fluid fæces and gas, resulting in an obvious and rapid relief of the gross distension and the patient's symptoms.

In 1 case about ten minutes after the contents of the loop had been emptied, a second rush of fluid suggested that the rest of the colon was now emptying by a mass peristaltic action. The lower end of the flatus tube was then (if the twist was high) connected with a second tube, so that as the sigmoidoscope was withdrawn, the tip of the flatus tube remained in situ above the level of the twist. The lower end was then stitched to the perineal skin, using local anæsthesia, and retained for 48 hours.

Difficulties and precautions: In a couple of instances the pelvirectal junction was drawn up so high that the sigmoidoscope did not quite reach the twist.* Here the tube was introduced gently in the direction seen and reduction took place. In 1 case the bowel at the base of the volvulus was drawn over to the left side so much that it was impossible mechanically to introduce the sigmoidoscope in the direction of the last few centimetres below the twist as the opposite tuber ischii obstructed the movement. It was therefore abandoned and laparotomy advised. The patient, however, refused, and went away unrelieved.

In 1 case congestion at the site of the twist was extreme, and after reduction doubt was felt as to the integrity of the bowel. It was therefore followed at once by a laparotomy; but this showed the tube in place and the volvulus relieved. A small patch of doubtful viability at the apex of the loop was invaginated.

Results: The manœuvre was used 14 times for volvulus, and succeeded in 13. One woman pulled out the tube twice (when it was only strapped) and then developed an inflammatory mass in the left iliac fossa but was not obstructed.

Before confidence was established in the conservative method of treatment, reduction by laparotomy and insertion of a flatus tube from below guided by a hand in the abdomen was used 4 times in the early part of the series in 2 patients (3 attacks; 1 attack). They both came back with recurrences later, which were then relieved by sigmoidoscope and tube.

2. *Curative Treatment*.—All the patients were advised to have a curative operation following the reduction to prevent recurrence, but only 1 of these (who had had three previous reductions) consented. It was carried out only 5 days after the third attack was relieved by sigmoidoscope and tube—in case he changed his mind and refused operation. Adequate bowel preparation was possible and the resection was carried out in one stage removing 14 in. of bowel and doing an end-to-end anastomosis. Clinical progress was satisfactory and this was confirmed by a later radiograph (which showed slight stenosis). He was discharged 16 days after admission.

The other patient treated curatively had had numerous attacks for two years with spontaneous relief, the last attack being in hospital. He still had signs of subacute obstruction so a laparotomy was performed. The pelvic colon was found to be narrowed at both ends and the pelvic loop was broadened till it resembled a stomach with a thick wall, and was filled with semi-solid fæces. The mesocolon was greatly thickened. A Paul-Mikulicz type of resection was performed in stages. Length of stay in hospital, 50 days.

B. Serious Type

Clinical Features.—The remaining 5 patients belonged to the group in which there was definite or suspected gangrene. Three of them came in a condition of extreme surgical shock and all of these had total gangrene of the pelvic loop.

The characteristics of this group were that it was their first attack, and they came to hospital *earlier* than the benign type, in 2 of the 3 cases.

These cases with gangrene all had very severe pain, marked diffuse tenderness, and a silent abdomen, with a weak and rapid pulse. One, a woman at full term, had an interlocking volvulus of the small intestine (which was resected) and of the pelvic colon (which was exteriorized.)

In the 2 other cases with total massive gangrene the whole loop was excised with closure of both ends through healthy bowel and a cæcostomy in 1 case and a transverse colostomy in the other. Neither survived.

The remaining 2 patients were less gravely ill, but gangrene was suspected because of a more rapid pulse than in the usual type and diffuse tenderness. At laparotomy, gangrene was not found and reduction was carried out by a rectal tube from below. One of these patients died on the third day (? peritonitis).

The incision in these cases was a practically total left paramedian to allow the gangrenous pelvic colon to be delivered without rupture.

Results of Treatment of 24 Attacks (18 Patients)

Treatment	*No.*	*Lived*	*Dead*	*Mortality per cent*	
Spontaneous reduction	1	1	—		
Non-operative reduction	14	13	1 (Ran away, presumed dead)	7	i.e., 10 per cent in all cases without gangrene
Operative treatment: Without gangrene	6	5	1	16	
With gangrene	3	—	3	100	
Total	24	19	5	21	

Discussion

Methods of Treatment and Mortality.—It is pointed out that the average mortality in volvulus is still about 45 per cent (Aird, 1941). Isaacson

* Probably a longer sigmoidoscope of 30 cm. with a diameter of about 2 cm. would aid in passing the twist under vision and reaching the higher torsion sites.

and Greer (1952), in discussing this, stated that it lay between 25 per cent and 60 per cent in series published in the U.S.A. The main reason for this high mortality lies probably in delayed diagnosis and treatment rather than in the method employed.

Treatment of the Attack—'benign' type.—The generally accepted treatment of the attack by laparotomy and reduction by a flatus tube passed per rectum generally gives good results for the occasional case if carried out early enough, yet it is pointed out that no more is achieved than by a conservative reduction as the predisposing long pelvic loop is not dealt with. Therefore, if as good results can be obtained by a conservative reduction which avoids the discomfort and hazards (even though slight) of an operation, it deserves favourable consideration.

The conservative method of reduction was introduced by Professor Hj. Schilling (Ulleval Hospital, Surgical Dept. III) about 1918. It has been the routine method of treatment in this hospital since (in the absence of symptoms and signs and gangrene), being carried out by a considerable number of surgical assistants. The collected results were reported by Bruusgaard (1947) and give impressive evidence of the safety of conservative reduction, showing a total mortality of 7·7 per cent in 168 treatments of all types, including resections (in 91 patients), and for 134 attacks treated conservatively, only 3 per cent. Accidental perforation occurred in 1 case (0·7 per cent).

Isaacson and Greer (1952) reduced 7 cases conservatively with 1 mortality.

The mortality in the present series of the selected group treated by conservative reduction was only 7 per cent, and after considering the patient who ran away as a fatality.

Probably the conservative method has its chief value in countries where volvulus is a relatively common condition and is well recognized. Here also the following safeguards are suggested—It should be carried out when :—

a. Pelvic colon volvulus is definitely diagnosed clinically and confirmed radiologically.

b. The clinical symptoms and signs indicate the 'benign' type with no evidence of gangrene or suspicion of threatening gangrene or peritonitis. Such a case will often, but not always, have a history of previous attacks.

c. The surgeon who performs it is prepared to proceed immediately or at any time within the next 48 hours to a laparotomy if not fully satisfied that reduction has been obtained without perforation.

Immediate radical treatment with resection of the pelvic loop in the absence of gangrene, and one-stage end-to-end anastomosis has been employed by Dean and Murray (1952), in 11 cases with 1 mortality (9 per cent). In spite of this good result it is generally accepted that primary resection and anastomosis is undesirable owing to the œdematous condition of the bowel above the volvulus. Alajem (1952) and others have employed routine exteriorization as a primary operation, proceeding to a Paul-Mikulicz resection. This, however, is a time-consuming method. Others (Handfield-Jones and Porritt, 1951) recommend colostomy and later extraperitoneal closure.

Curative treatment: Neither plication of the mesocolon (which is already stiff, and thickened usually) nor colopexy (which is dangerous in the acute phase) has been attempted in this series to prevent recurrences, as they appear to be mechanically inadequate.

If the comparative safety of a conservative reduction in the absence of gangrene is accepted, then the best method of treatment would appear to be a conservative reduction of the attack with the precautions suggested, followed by a one-stage resection at an interval of 7–10 days, giving time for adequate bowel preparation and a return of the local circulation to normal.

A one-stage resection, now a generally accepted procedure, was recorded already in 1939 (Thompson).

Treatment of Pelvic Volvulus with Gangrene.—The simplest treatment is exteriorization of the gangrenous loop followed by a Paul-Mikulicz resection. However, this carries a definite risk of gangrene of the *lower stump*, owing to thrombosis of the sigmoid branches of the inferior mesenteric artery. This gangrene was not present at the time of exteriorization in one case of this series, but appeared after some days, possibly owing to an extension upward of the thrombosis so that the superior hæmorrhoidal artery also became involved. The patient developed fatal peritonitis following the gangrene.

In a case of Semb's (1955) the patient developed a local abscess around the lower stump, and following a second operation to restore continuity this abscess resulted in a fatal peritonitis.

Subsequent cases in this series were therefore treated by a closure of both ends of the pelvic loop after resection, making sure that this was carried out through well-nourished bowel wall, with a proximal colostomy preferably of the right transverse colon. In successful cases restoration of continuity in a clean field should then be possible after an interval.

Semb has adopted the method of closure of the distal end after resection, and bringing out the proximal end as an end colostomy. Where the patient's condition is grave this would be preferable as taking less time.

Resection of the gangrenous loop should be carried out *without detorsion* to avoid the toxic products in the imprisoned circulation of the loop entering the general circulation (Stevenson, 1955).

SMALL INTESTINE VOLVULUS

History.—The duration of symptoms before coming to the hospital varied between 20 hours and 12 days, the average being about 3½ days. There was a severe constant pain in most cases but also in others colicky pain, and vomiting was often persistent. They complained less of the constipation and distension than of the abdominal pain.

Physical Examination.—The pulse was usually 100 or higher. Usually afebrile. The abdomen was moderately distended, usually soft, and in several cases definite peristalsis of small-intestine pattern was visible associated with high-pitched sounds on auscultation. Tenderness was present in the right iliac fossa on several occasions, and in some cases generalized tenderness and guarding. Ascites could be detected in the majority. In 3

cases which proved to have massive gangrene, the general condition was bad, the abdomen silent or with only occasional peristaltic sounds.

X-ray Findings.—A plain radiograph was taken soon after admission with the patient standing. The appearance gave definite evidence of small-intestine obstruction, with inverted distended loops containing fluid levels, but differed in several respects from a simple non-strangulating obstruction (when the inverted loops arise *regularly* one above the other from the right-iliac fossa to the left sub-costal region, in relation to the attachment of the mesentery) as follows :—

1. The loops, which may show valvulæ conniventes, were wider and less regularly placed and less active (activity being judged by the difference in level of the fluid in the two limbs of a loop)—indicating obstruction with reflex partial paresis of the coils involved and above the volvulus.

There was frequently marked ascites which to some extent obscured the picture.

3. Occasionally a loop or loops can be almost circular and pointing to the right iliac fossa—indicating the actual loop involved. This was observed once only.

4. In some cases there was very little gas and a few small fluid levels only. This suggests a tight twist not allowing the gas to pass into the involved segments and paresis above and hence indicates an increased liability to early gangrene. (Frimann Dahl.)

While the findings were not so obvious as in the large-bowel volvulus, and could not be distinguished from internal strangulation due to causes other than volvulus, they indicated clearly the need for urgent laparotomy.

Treatment.—

Pre-operative.—Operation was delayed 2–4 hours while as far as possible dehydration was relieved by intravenous fluids ; the stomach was emptied by a Ryle's tube which was kept in situ, and continuous or intermittent suction maintained throughout operation.

Operation.—A generous right mid and lower paramedian incision was used under general inhalation anæsthesia. On opening the peritoneum excess of free fluid was invariably present and it was blood-stained in about 50 per cent of the cases. The distended darkly congested coils of small bowel were very obvious and also the pale collapsed cæcum, which was drawn medially.

The first step was to pass a hand between the distended bowel and the parietal peritoneum to feel for, and if present divide, any band following inflammatory conditions of the parietes that may have predisposed to the condition, or a congenital band, e.g., from a Meckel's diverticulum, passing to the parietes.

Closure was assisted by intravenous myanesin, 10.0 c.c., to give relaxation if necessary. The bowel was not aspirated.

In 3 patients there was massive gangrene, of about 10–11 ft. in one case and 2½ ft. in the others. Resection was carried out. In one case entero-enteric anastomosis was carried out ; in one, owing to the proximity of the gangrene to the cæcum, entero-colic anastomosis ; and exteriorization of the loop in the third because of very bad general condition.

Post-operatively gastric suction by Wangensteen's method and intravenous fluids were given for about 48 hours. Fluids by mouth were given as soon as possible, and the suction was used intermittently till it could be safely removed. Fluid charts were maintained, but biochemical investigations were not available. Antibiotics and the sulphonamide group of drugs were given for some days. Blood was not generally available for transfusion. In the later cases, when the bowel was healthy, prostigmine was given 4-hourly till the abdomen was no longer distended, and possibly contributed to improved results.

Even apart from gangrene the mortality-rate was high. However, the last 3 cases which came within a week and lay side by side in the ward all recovered.

The results in the small-intestine series of 14 cases was as follows :—

Treatment	*No.*	*Lived*	*Died*	*Mortality per cent*
Laparotomy and simple untwisting	9	5	4	44
With gangrene :—				
a. Suture of local gangrene line	2	1	1	50
b. Resection	3	—	3	100
	14	6	8	57

Next the congested and distended small bowel involved was fully exteriorized. It was found with experience that it was more dangerous to attempt reduction without exteriorization as gentle movements were essential and also room in which to rotate the bowel mass freely. The exteriorized coils were covered in large saline wraps, supported by the assistant.

The right hand of the surgeon was then placed at the root of the volvulus in the precæcal region. The direction of the twist could not generally be observed by inspection. With the help of the assistant the whole involved loop was then twisted gently, first in one direction and if necessary in the other, and the hand at the root could then definitely detect if the root became tighter or looser. As untwisting continued the 'knot' at the base became looser till it was possible to pass the hand unhindered from the root of the mesentery along the mesentery surface to the bowel. Extreme caution was required. because there is a liability to local necrosis due to the pressure on the anterior wall of the bowel loop lying posteriorly at the torsion site. On one occasion this ruptured, and was repaired by two layers of sutures ; in another the linear necrosis was invaginated by a few interrupted catgut sutures.

The effect of untwisting was dramatic, with a return of normal colour (except where gross gangrene had occurred), and the cæcum could be observed to fill with gas, which also passed into the rest of the colon. In the later cases at this stage a flatus tube was passed per rectum and prostigmine given (1·0 c.c. I.M.) (when the bowel wall was healthy in appearance and reacted to gentle pinching) and a satisfactory passage of gas and some fluid per rectum was noted. The importance of not allowing the

mass of small-intestine coils weighted with fluid to fall by gravity to one or other side was realized by the evident dark-blue congestion when this occurred.

Discussion

Methods of Treatment and Mortality.—There is no alternative in this group to an urgent laparotomy, and this should be carried out also in cases in which the diagnosis is in doubt. The use of the Miller-Abbott tube has no place in the treatment of small-intestine volvulus because of the increasing risk with the passage of *hours* of both general loop and local pressure gangrene. In spite of laparotomy without delay, however, the mortality in this series is very high.

It is 44 per cent for cases without gangrene, while that of the similar cases in the pelvic colon group is only 10 per cent. This points at once the major difference in prognosis between the two types, which is probably due to the factors of fluid and electrolytic loss and absorption of toxic peritoneal exudate already mentioned.

The absorption of toxic bowel contents after reduction may also be important. The coils at operation are heavy with fluid, which probably amounts to at least 5–6 pints. After untwisting and restoration of the normal circulation, a rapid increase of the abnormally toxic bowel contents may take place, with a resulting circulatory collapse (Aird, 1941).

It is therefore probably important to empty the small intestine rapidly to avoid this risk as much as possible. Kerr and Kirkaldy Willis (1946) achieved this by a direct opening of the bowel and emptying it into a sterile bowl.

In the last 4 cases of this series a flatus tube was passed per rectum and 1·0 c.c. prostigmine given (the bowel wall being healthy) and gas and fluid were observed to pass on the table, with evident relief of the great distension.

This certainly also had the important effect of making closure easier and safer. All these four cases recovered. If further experience with this method confirms these good results, it would appear to be preferable to methods which involve an opening in the damaged bowel wall.

Relationship of Mortality to Time elapsed between Onset and Treatment.—Michael (1940) points out a clear relationship between the time that elapses between the onset of small-intestine obstruction and treatment. In his series the average time was 2 days in 23 cases that survived and 6 days in 12 fatal cases. While this relationship is of the greatest importance, and late arrival at hospital probably accounts in part for the high mortality of this series, yet the present series does not show that cases that came early after the onset have necessarily a better prognosis. In fact a high proportion of the cases with gross gangrene affecting the whole of the loop or loops involved, both in the small- and large-intestine series, had a very short history, and came quickly to the hospital just because of the dramatic severity of the symptoms.

The relationship observed for both small-intestine and pelvic colon volvulus is recorded graphically in *Fig.* 176.

Summary of Results of Treatment in All Types

Site	No. of Patients	No. of Attacks	Treatment	Lived	Died	Mortality
						per cent
Cæcum	1	2	Operation	1		
			Spontaneous	1		
Small intestine	14	14	Operation	6	8	57
Pelvic colon	18	24	Spontaneous (1)	1		
			Non-operative (14)	13	1	7
			Operative (9)	5	4	44
Total	33	40	—	27	13	32·5

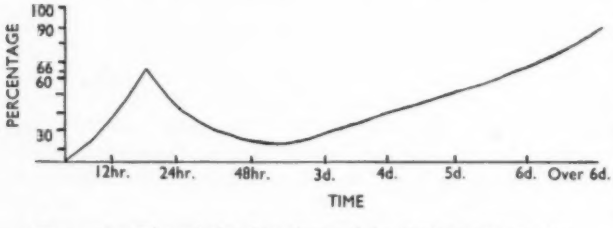


Fig. 176.—The relationship observed for both small intestine and pelvic colon volvulus.

Volvulus of the Cæcum

The single case seen recovered after laparotomy and untwisting. The cæcum was not fixed and she returned with a recurrence of symptoms, which subsided spontaneously, after 5 months. This single case will not be further considered.

Summary

1. A series of 40 attacks of volvulus occurring in 33 patients in Western India is presented.

2. The incidence of volvulus and some of its varieties in different countries is considered.

3. The relative importance of inflammatory adhesions, diet, and congenital anomalies is discussed, and secondary changes tending to recurrence are described.

4. The common clinical pictures are presented with an emphasis on differential diagnosis between the small-intestine and large-intestine varieties.

5. The radiological findings are given, and their importance emphasized in diagnosis.

6. Treatment of volvulus of the pelvic colon by a conservative method is described and recommended, subject to adequate precautions, followed by a one-stage resection of the redundant pelvic loop after a short interval.

7. The importance of rapid emptying of the small intestine, after reduction of the volvulus by urgent laparotomy, is stressed, and a method recommended for further trial.

8. The mortality in relation to different methods of treatment is discussed briefly.

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SUPRACONDYLAR FRACTURES OF THE HUMERUS IN CHILDREN

A REVIEW OF 100 CASES

By W. LAURENCE

BRIGHTON AND LEWES GROUP OF HOSPITALS

"THE supracondylar or transcondylar fracture is above the elbow-joint. If the surgeon does not manhandle the elbow-joint with repeated forcible manipulations, or insult it by open surgery, there will be useful elbow motion. There may be deformity, there may be limitation, but there will be useful elbow-joint motion."—Vernon P. Thompson, 1951.

This quotation from a discussion on fractures of the elbow in children is borne out by my observations in a review of 100 cases of supracondylar fracture. The main feature which contributes towards a satisfactory result in any severe supracondylar fracture is the capacity of the bones to undergo remodelling after displacement of the fragments. Attenborough (1953) showed in his four cases, chosen to show the results of failed reduction, the good results following remodelling.

All the supracondylar fractures treated in this unit between 1950–4—a total of 100 cases—were reviewed with particular reference to the outcome after displaced fractures, and the incidence of complications. The average age of the children was 6·5 years, the youngest being 2 years old, and oldest 11 years. The average time required for union was three weeks, and for restoration of full movements eight to twelve weeks. There were 3 cases of transcondylar fracture.

REDUCTION AND IMMOBILIZATION

Manipulative reduction was carried out under general anæsthesia with serial radiographic control. Only when casualty house-surgeons had been trained in the technique were they allowed to treat these cases. The manœuvre of traction with flexion was used, and the elbow was immobilized by a posterior slab and strut, the slab being held in place by cotton bandages. This allowed maximal swelling without risk of circulatory impairment. The children were admitted for elevation of the limb and observation.

Since there is a tendency for the fracture to slip up to the fifth day, a check radiograph was taken then. If the position remained satisfactory the plaster was completed, and the patient discharged from hospital. If the fracture was found to have slipped, reduction

FIG. 177.—Anterior beak present.

FIG. 179.—*Case* 1. Remodelling in progress, note marked posterior 're-shafting'.

was undertaken again. As a rule no more than two reductions were carried out, and if the fracture slipped a third time the position was accepted.

COMPLICATIONS

Impairment of Radial Pulse.—In 6 of the cases the radial pulse disappeared after reduction of the fracture, but the circulation to the fingers was clinically satisfactory, and the patients did not complain of pain. These cases were treated conservatively by slitting the cotton bandage in the whole of its extent, and elevating the arm. The amount of flexion at the elbow was not altered. The pulse returned about one week later, when the swelling had subsided. In no case did an ischæmic lesion develop.

Median Nerve Lesion.—In 4 cases the median nerve was clinically involved, as manifested mainly by sensory symptoms and signs. In 2 of the cases muscular signs also existed. The sensory changes were those of diminished sensation, mainly to tip of the index finger, which in all 4 cases became pointed. This is a feature I have not seen mentioned before. The motor changes consisted in wasting of the thenar eminence, with weak response to faradic current.

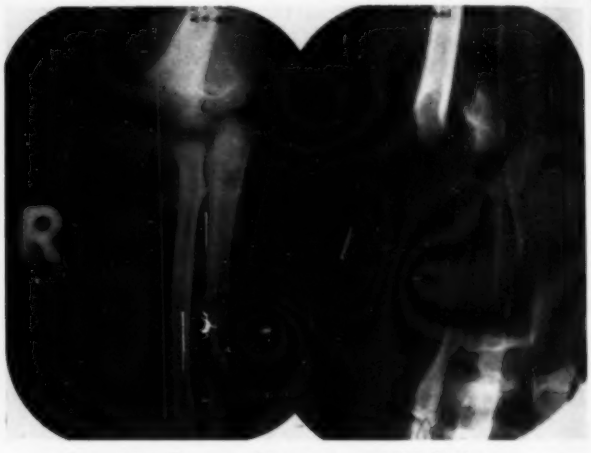

FIG. 178.—*Case* 1. Gross posteromedial displacement.

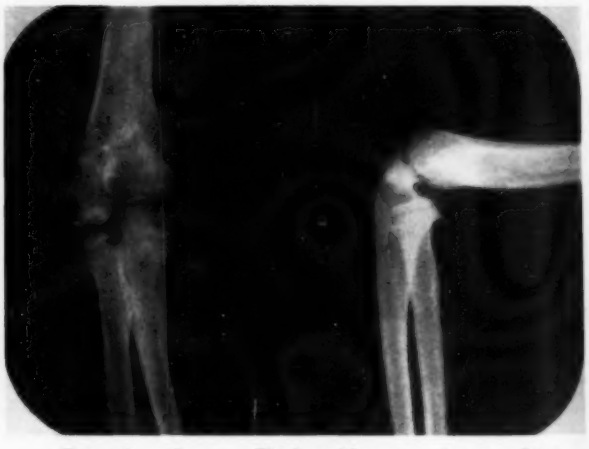

FIG. 180.—*Case* 1. Final architecture 3½ years later. There is gross posterior displacement, which was eventually remodelled, until 3½ years later there was a normal appearance with full movements.

Two of the cases were associated with beaking (*Fig.* 177), the third with a loose anterior fragment, and the other with forward tilt of the lower fragment. These deformities were also present in a number of uncomplicated cases, and it is not suggested that the association is causative. In the 4 cases with median nerve impairment motor and sensory recovery was complete in seven or eight months.

REMODELLING AFTER DISPLACED FRACTURES

It seems clear that bone remodelling can overcome a fair amount of backward displacement, and that a full range of movement will eventually be restored; but increase in flexion beyond the normal tends to leave a corresponding limitation in the last few degrees of extension. Conversely, reduction of the lower fragment to aline with the upper fragment, and without normal forward tilt, will result in the fracture slipping when the normal amount of fracture line absorption occurs. The lower fragment should be flexed to 20–25°. Radiographs illustrating the process of remodelling are shown in *Figs.* 177–189.

FIG. 181.—*Case* 2. Fracture displaced in plaster.

FIG. 182.—*Case* 2. Displacement still evident, but re-shafting at four weeks.

It is notable that in none of these examples was a valgus or a varus angulation corrected by remodelling. It was felt that if the anteroposterior displacement could not be corrected, then at least the lateral angulation could. Once an excess of valgus or varus is initiated it tends to be permanent, for its correction appears to be beyond the remodelling powers of the periosteum. It seems that such angulation is due to gravity rotating the lower fragment into pronation (Boyd, 1944).

Excessive Flexion.—As pointed out by Watson-Jones (1943), the normal forward tilt of the lower humeral epiphysis is 25°. If, after a supracondylar fracture, this amount is increased, extension is limited to a corresponding degree (*Fig.* 190).

Cubitus Varus.—In every case, except one, in which the carrying angle was altered the change was towards cubitus varus. Correction was only undertaken in this case, that of a girl in whom the 'crooked' elbow was unsightly.

Cubitus Valgus.—This should be corrected by lateral linear greenstick osteotomy to prevent the risk of delayed ulnar palsy.

DISCUSSION

Boyd (1944) noted median nerve lesions in 1·5 per cent of his cases, and radial nerve lesions in 2·4 per cent. Ulnar nerve lesions occurred twice; but since the figures do not indicate the total number of patients under review their incidence is not known. In my series there were 4 cases of median nerve involvement, but there was no instance of radial or ulnar nerve involvement. I believe that impairment of the median nerve is caused by a mild ischæmic lesion, for it accords with the usual observation that ischæmic lesions are either sensory, or combined motor and sensory, never motor alone. The fact that these fractures do interfere with the circulation to a greater or lesser extent also supports the hypothesis of ischæmia. If the lesion were a nerve lesion in continuity, such as follows severe contusion, then complete motor as well as sensory changes should be present. It is difficult to see how the ulnar nerve could be involved either by contusion or ischæmia.

FIG. 183.—*Case* 2. Final architecture one year later. Following a satisfactory reduction the fracture slipped. The final result was an elbow with normal carrying angle and full movements.

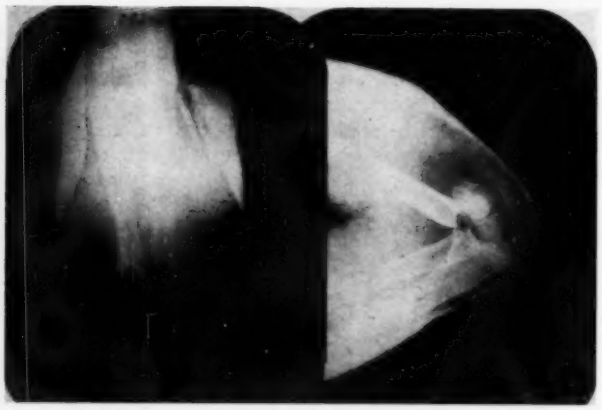

FIG. 184.—*Case* 3. Complete posterior displacement. No lateral displacement.

FIG. 185.—*Case* 3. Remodelling six weeks later.

FIG. 186.—*Case* 3. Finally at 4½ months. Fracture reduced twice, but slipped. There was marked posterior, but no lateral, displacement in accepted position. This caused a 'beaking' of the shaft which did not interfere with movement as it grew away from the joint.

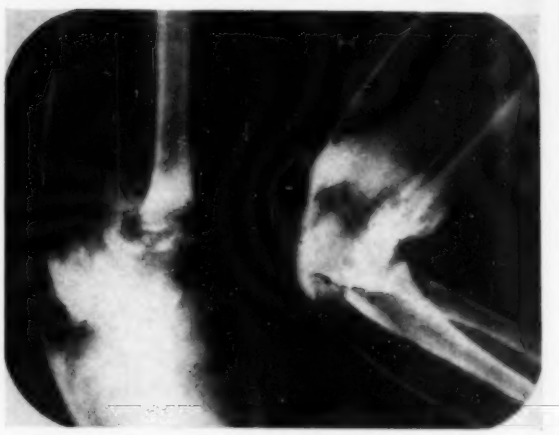

FIG. 187.—*Case* 4. The accepted position following the third manipulation.

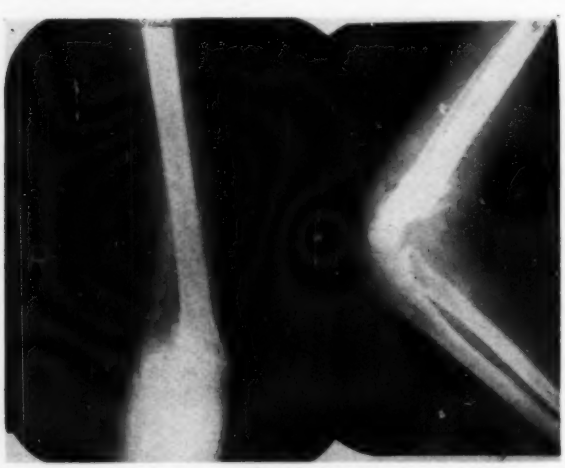

FIG. 188.—*Case* 4. Remodelling at six weeks.

FIG. 189.—*Case* 4. Three years later. This case illustrates a complete remodelling sequence. Reduction was carried out three times, but final position was one of displacement. The radiographs show the stages to a normal architectural conclusion.

Boyd recommended that the arm should be put on traction with the elbow slightly flexed, if there was much swelling, and that manipulation should be deferred. He advised a 'partial reduction' if there was interference with the pulse. Should closed reduction fail, he recommended open operation to fix the fragments with cross-nails. Surely, if reduction is undertaken, it should be as complete as possible, for the loss of pulse may be due to spasm, and local pressure cannot be eliminated until the fracture is reduced.

Blount, Schulz, and Cassidy (1951) also advocated traction in the presence of gross swelling, with manipulation of the fracture ten days later. About

circulatory disturbance he wrote : " An absent radial pulse *per se* is not an indication for open operation." He agreed that the most important herald of ischæmia is pain.

Thomas (1950) placed more reliance on the colour of the fingers than on the radial pulse. I have found

FIG. 190.—*Case* 5. Increase in forward tilt, limiting extension. The accepted reduction has a forward tilt of 60°. This resulted in loss of last 25° extension.

that this is justified, and that the colour is a reliable guide to the state of the circulation.

Cox, Hurley, and Compton (1952) noted that too much flexion of the lower fragment led to limitation of extension. They also stated, without reservation, that incomplete reduction will not be corrected by remodelling.

SUMMARY AND CONCLUSIONS

1. Supracondylar fractures are easy to reduce provided reduction is controlled by serial radiographs and the patient is not struggling under inadequate anæsthesia. The initial reduction is not always easy to maintain. It may not be safe, because of swelling, to flex the elbow sufficiently to be sure of stability.

2. Reduction must be carried out as an emergency before gross swelling occurs. If reduction is delayed and severe swelling has occurred, elevation by slinging in plaster is recommended.

3. When redisplacement occurs it is often at about the fifth day ; so a routine check radiograph should be taken then. The more swollen the elbow at time of reduction, the more likely are the fragments to slip.

4. Manipulation should be repeated if necessary, but risk of circulatory disturbance does not warrant more than two attempts at reduction.

5. If the backward displacement cannot be corrected the normal carrying angle should be restored.

6. Because of the capacity of the bones to remodel themselves it is probably never necessary to undertake open reduction for the fracture itself.

7. A median nerve lesion complicating a supracondylar fracture has a good prognosis for full recovery.

8. It is suggested that reduction is a skilled manœuvre, which should not be delegated to a casualty officer until he has received adequate supervised instruction.

9. A plaster slab and strut helps to maintain position, enables the arm to be slung, and diminishes pain.

10. If the radial pulse has been felt immediately after reduction, but disappears later, there does not appear to be a serious risk of ischæmic complications, provided there is no constriction at any point of the extremity.

I wish to express my thanks to Mr. Lloyd Williamson, Mr. R. Brooke, and Mr. J. Crawford Adams for their help in the preparation of this paper.

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RIGHT HAND, LEFT HAND: THE SURGICAL APPROACH TO THE BILIARY PASSAGES

BY H. A. HAXTON AND E. A. MCVERRY, MANCHESTER

THE usual approach to the bile-ducts and gall-bladder is through one of a variety of incisions on the right side of the abdomen, the surgeon standing on the patient's right side. Four years ago one of us (H. A. H.) began to operate from the left side, using a median epigastric incision, and since then more than 200 operations on the gall-bladder and bile-ducts have been approached in this way in patients under our care. We feel that the method has such definite advantages over the traditional routes that a detailed account of the procedure is desirable.

METHOD

The surgeon stands on the left side of the patient throughout the operation, and in all cases a midline incision is used. It has never been found necessary to extend the incision below the umbilicus.

The abdomen open, a general survey is carried out, any adhesions around the gall-bladder are divided, and the biliary tract is palpated. To do this latter the surgeon places his left hand so that the index and middle fingers are inserted into the foramen of Winslow behind the common bile-duct and the thumb lies in front of it. Sensitive palpation

of the duct area is now possible. If any suspicious thickening is felt in the head of the pancreas the peritoneal reflection lateral to the second part of the duodenum is easily broken through with the fingers of the left hand, which are then passed behind the duodenum and head of the pancreas. Accurate palpation of the lower part of the common bile-duct between the thumb and fingers can be carried out,

FIG. 191.—Insertion of the left index finger through the 'cysto-hepatic triangle'.

and as a rule any gall-stones impacted at the lower end of the duct can be squeezed into its supra-duodenal portion.

If cholecystectomy is decided upon a pack is placed behind the surgeon's left hand. This controls the stomach and intestines and it also imposes a downward pressure on the duodenum, which places the common bile-duct on stretch. A Deaver retractor is placed in the upper end of the wound so as to overlie the quadrate lobe of the liver and to hold the falciform ligament out of the line of vision. This retractor is held in such a position that the tip does not depress the liver. It has rarely been found necessary to use further retractors.

The peritoneum is incised over the cystic duct and the surrounding fatty tissue is dissected away. The fingers of the left hand brace the duct forwards and spread the tissues so as to facilitate accurate observation and dissection. The cysto-hepatic triangle is rendered visible, especially in asthenic patients, and the left index or middle finger may be hooked forward between the neck of the gall-bladder and the liver (*Fig.* 191). This manœuvre facilitates the definition of the cystic vessels and duct. It will be appreciated that throughout these procedures the structures are being palpated continuously and placed on stretch at will. As a result any abnormalities of the vessels or ducts are detected more readily and the risk of damage to important structures is reduced.

Straight artery forceps are applied to the cystic artery and duct, since we have found that the usual curved clamps make the application of ligatures more difficult from the left side. After ligation and division of these structures the gall-bladder is carefully freed from the liver by scissors or finger dissection. The peritoneal reflections are left long when possible and no suturing of these or of the gall-bladder bed is done as it is not considered to be advantageous.

Should it be considered advisable to open the common bile-duct there is no need to use stay sutures, as the duct can be held forward on the stretch by the fingers of the left hand which lie behind it. Furthermore, it is much easier to detect and to milk out stones both small and large from the duct with the fingers in this position. If the bile-duct and bile are relatively normal we suture the opening in the duct and insert a drain down to, but not into, it.

Appendicectomy if indicated is usually straightforward, ample exposure being obtained by elevating the abdominal wall with the Deaver retractor placed at the lower end of the wound.

RESULTS

For the purpose of comparison 100 patients with cholecystectomy were examined and reviewed three months to two years after operation. Fifty had been operated on from the left side by median wounds and 50 from the right side by paramedian wounds.

One death occurred in each series, the causes being cardiac failure on the second post-operative day in one and pulmonary embolism on the eighth day in the other.

It can be seen (*Table I*) that while there is no striking difference between the two series the patients with midline wounds tended to have shorter wounds and also a rather shorter stay in hospital. In the midline group one incisional hernia occurred; in the paramedian group there were 2 incisional hernias and 1 case of wound dehiscence. These findings suggest that the soundness of the midline wound is not inferior to that of the paramedian. Ward sisters have commented, and it has been our own impression, that patients with midline incisions had less pain and discomfort in the early post-operative period.

DISCUSSION

We make no claim for originality in the procedure described, for we are aware that the left-sided approach is used by several surgeons in this country and that it has been used and advocated for over twenty years in Italy by Professor Pettinari (1952). Nevertheless, the method is not widely used, as was shown by the replies to a questionnaire sent to 100 surgeons in U.S.A. and reported in an editorial article in the *American Journal of Surgery* (1951). Every one of the 97 right-handed surgeons replied that he stood on the patient's right side throughout the operation. The same article advocated a right paramedian incision made from the right side followed by the surgeon moving round to the patient's

left side. It was stressed that the placing and tying of ligatures is easier from this side. Similarly, Maes and Weiss (1951) recommended moving round to the left side to palpate the common bile-duct. This changing of sides appears unnecessary since the whole operation can be done with advantage from the left side.

Despite this lack of popularity, we and others who have worked with us are convinced that considerable advantages accrue from the use of the left-sided approach through a median incision. This incision is quickly made and bleeding from it is minimal. It is situated entirely above the umbilicus and so there is no tendency for transverse colon, omentum, or small intestine to be extruded during the operation or to become adherent to the scar during the period of healing. The median incision is more quickly and easily closed than the paramedian one, especially in obese patients. In these the readiness with which sutures cut out of the posterior rectus sheath during the repair of paramedian wounds is a well-known source of annoyance to surgeons and the cause of many incisional hernias.

Table I.—Comparison of Cases with Cholecystectomy through Median and Paramedian Wounds

Incision	No. of Cases	Average Age (years)	Male	Female	Average Stay Post-operative (days)	Wound Dehiscence	Scar Length	Hernia	Died
Midline	50	51·7	6	44	15·5	0	6·0	1	1
Right upper paramedian	50	53·0	2	48	16·1	1	7·4	2	1

Standing on the left side, the surgeon has a direct view of the gall-bladder and bile-ducts, which, lying to the right of the midline, tend to be hidden by the right edge of the wound when the surgeon stands on the patient's right side. Furthermore, a large stone in Hartmann's pouch or a distended gall-bladder can obstruct the view, from the right side, of the bile-ducts, necessitating traction by forceps on, or aspiration of, the gall-bladder. The view from the left side is not obstructed and with this approach we have never found it necessary either to apply forceps to the gall-bladder or to aspirate it when distended. Avoidance of these steps must reduce the risk of peritoneal contamination.

From the left side, too, the surgeon can easily and naturally use his left hand to palpate and spread out the parts of the biliary tree. This is in striking contrast to the awkward angle at which the left hand is inserted when operating from the right side and which impairs the usefulness of the left hand during the operation.

As a result of the improved visualization from the left side we have found it much easier to ligate the cystic artery separately from the duct than we did from the right side. Should bleeding occur from the cystic artery the surgeon on the left side is much better placed to control it, for he can easily compress the hepatic artery between the left thumb in front and the index and middle fingers behind in the foramen of Winslow. Allen (1945) advocates this manœuvre, but if the surgeon is operating from the right side it necessitates his going round to the left side of the patient, so wasting valuable time and blood. Controlling the blood-flow in this way it is easy to pick up the bleeding point exactly. This minimizes the risk of damage to the common bile-duct, for it is generally agreed that damage to this structure often results from efforts to control bleeding from the cystic artery.

The other common cause of injury to the main bile-duct is traction on the gall-bladder and cystic duct during ligation of the latter, resulting in tenting of the bile-duct and the application of the ligature or forceps to its wall. Since traction is unnecessary for good visualization from the left side, and since the ducts can be spread out on the fingers of the left hand, the possibility of such an injury is greatly reduced in our opinion.

SUMMARY

The approach to the surgery of the gall-bladder and bile-ducts from the patient's left side via a median epigastric incision is described.

From our experience of this left-sided approach we conclude that it makes biliary surgery easier and safer by enabling the surgeon to see and feel the ducts and vessels more readily.

The median incision is made and closed more quickly than the paramedian, and our follow-up of 50 cases done by each type of incision has indicated fewer wound complications in cases with median wounds.

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ANÆMIA FOLLOWING PARTIAL GASTRECTOMY

A REVIEW OF 100 CASES

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ANÆMIA is regarded as a common occurrence following partial gastrectomy; it may be relatively symptomless and only discovered during routine follow-up examinations.

It is commonly stated that a hypochromic and microcytic anæmia is the usual type to be found following partial gastrectomy. Gordon-Taylor, Hudson, Dodds, Warner, and Whitby (1929) found some degree of hypochromic anæmia in 23 out of 52 patients. Hartfall (1934) studied 40 cases of anæmia following gastrectomy and gastro-enterostomy, and found a hypochromic and microcytic anæmia in all except 3 cases. Morley and Roberts (1928) found a hypochromic anæmia in 16 out of 42 cases.

On the other hand, Brain (1951) did not find any iron-deficiency anæmia in his cases, except in the immediate post-operative period, which he regarded as being caused by direct blood-loss before and at operation, with consequent depletion of iron stores. There was, however, a macrocytosis in 11 out of 35 of his cases, but none of his patients had an associated anæmia which required treatment.

Reports of cases of macrocytic anæmia with a megaloblastic bone-marrow occur from time to time in the literature, but are not a common finding. Larsen (1952) reported 3 such cases, and Hurst (1932) reported 5 cases following partial gastrectomy.

Lake (1928), however, found no anæmia in a series of 52 cases following partial gastrectomy.

PRESENT INVESTIGATION

In this series of 100 cases, of which 70 were men and 30 women, the average time of examination following the partial gastrectomy was 4 years 3 months, the single earliest case being 5 months after the operation and the longest time being 10 years. In the cases regarded as having normal blood-counts, 2 patients had been operated on 10 years previously and 1 case $7\frac{1}{2}$ years previously.

Of the cases operated on 78 were for gastric ulcer, of which 26 were associated with a duodenal ulcer, and 21 cases were for duodenal ulcer alone, the remaining 1 case being operated on for repeated hæmatemesis, no ulcer being found. A patient was considered to have an anæmia if the hæmoglobin level was 12·6 g. per 100 ml. (= 85 per cent Haldane) or less. This level was chosen because it was considered that many people, particularly women, have a hæmoglobin level between 12·6 g. and 13·4 g. per 100 ml. and are in perfect health.

In all the 100 cases a full blood-count and absolute values were determined, the normal mean corpuscular volume (M.C.V.) being taken as 78–95 cu. μ, and the lower limit of normal for the mean corpuscular hæmoglobin concentration (M.C.H.C.) 32 per cent.

Bone-marrow examinations were made when the anæmia was macrocytic in type.

Findings.—Of the 100 cases, 56, comprising 46 men and 10 women, were regarded as normal. The remaining 44 cases showed an anæmia of the following types :—

Type of Anæmia	*Number of Cases*	*Men*	*Women*
Normochromic and normocytic	14	5	9
Hypochromic and normocytic	19	13	6
Hypochromic and microcytic	9	6	3
Macrocytic	2	0	2
Total	44	24	20

In 14 patients, 5 men and 9 women, there was a normochromic and normocytic anæmia. This type of anæmia was found to be not severe. (*Table I.*)

Table I.—NORMOCHROMIC AND NORMOCYTIC ANÆMIA

CASE NUMBER	HÆMOGLOBIN	RED CELLS	P.C.V.	M.C.V.	M.C.H.C.
	g. per 100 ml.	million per c.mm.	per cent	cu. μ.	per cent
1. G.U. and D.U.	12·3	4·0	37	93	32·5
2. G.U.	12·6	4·6	40	87	32
3. G.U..and D.U.	12·0	4·2	38	91	32
4. G.U.	12·0	4·3	38	89	32
5. D.U.	12·0	4·3	38	89	32
6. G.U.	12·3	4·2	39	94	32
7. G.U.	12·0	3·9	35	90	34
8. G.U.	12·2	4·2	38	91	32
9. G.U. and D.U.	12·2	4·3	37	86	33
10. Hæmatemesis (no ulcer)	12·6	4·3	39	91	33
11. G.U.	11·7	3·9	37	94	32
12. D.U.	12·6	4·5	40	89	32
13. G.U.	11·7	4·6	37	80	32
14. D.U.	12·6	4·5	40	89	32

G.U.—Gastric ulcer. D.U.—Duodenal ulcer.

The majority of cases of anæmia were found to be hypochromic, but normocytic, i.e., a M.C.V. between 78–95 cu. μ, but having a M.C.H.C. of less than 32 per cent (*Table II*).

Only 9 cases had both a hypochromic and microcytic anæmia (*Table III*).

It will be seen that iron deficiency was present in 28 cases out of the total 44 cases of anæmia.

Of the 2 cases of macrocytic anæmia, only 1 case had a megaloblastic marrow. This patient, aged 69 years, showed the following blood-picture 15 months following partial gastrectomy (Billroth I) for a chronic gastric ulcer: Hæmoglobin 11·3 g. per

100 ml.; red cells 2·9 million per c.mm.; P.C.V. 34 per cent; M.C.V. 117 cu. μ.; M.C.H.C. 33 per cent. The sternal marrow examination showed the presence of megaloblasts, and the blood-count returned to normal on vitamin B_{12} therapy. As the patient's blood-picture was normal prior to operation, it is reasonable to assume that the macrocytic anæmia and megaloblastic marrow were due to the gastrectomy, and not to the development of Addisonian pernicious anæmia.

The second patient (aged 65 years) showing a macrocytic anæmia 3½ years following partial gastrectomy (Billroth I), had a count as follows: Hæmoglobin 12·3 g. per 100 ml.; red cells 3·0 million per c.mm.; P.C.V. 37 per cent; M.C.V. 123 cu. μ.; M.C.H.C. 33 per cent. The sternal marrow examination showed mild normoblastic

Table II.—HYPOCHROMIC AND NORMOCYTIC ANÆMIA

CASE NUMBER	HÆMOGLOBIN	RED CELLS	P.C.V.	M.C.V.	M.C.H.C.
	g. per 100 ml.	million per c.mm.	per cent	cu. μ.	per cent
15. D.U.	9·4	3·7	34	92	28
16. G.U.	10·7	4·0	38	94	29
17. G.U. and D.U.	9·1	4·0	33	83	28
18. D.U.	10·1	3·8	35	94	29
19. G.U. and D.U.	11·7	4·7	41	87	29
20. G.U.	12·0	4·5	40	89	30
21. G.U. and D.U.	10·4	4·2	36	86	29
22. G.U. and D.U.	10·4	4·2	36	86	29
23. G.U.	11·7	4·4	41	93	29
24. G.U.	10·1	4·0	37	93	28
25. G.U. and D.U.	8·8	3·4	32	94	28
26. G.U.	9·4	3·7	34	92	28
27. G.U.	11·3	4·4	37	85	31
28. G.U.	8·8	3·6	28	78	31
29. G.U. and D.U.	9·1	3·5	31	89	30
30. G.U. and D.U.	7·9	3·3	30	90	27
31. G.U.	10·4	4·0	37	93	28
32. G.U.	11·7	4·5	38	85	31
33. G.U.	11·3	4·5	36	80	31

G.U.—Gastric ulcer. D.U.—Duodenal ulcer.

hyperplasia, with some macronormoblasts, but megaloblasts were not seen. Her blood-count 5 months later was: Hæmoglobin 12·6 g. per 100 ml.; red cells 3·9 million per c.mm.; M.C.V. = 103 cu. μ.; M.C.H.C. 32 per cent. Unfortunately, it was not possible to keep this patient under review.

DISCUSSION

The causes of anæmia are variable; gastrectomy removes the greater part—if not all—of the hydrochloric acid-producing tissue, with the result that iron may be poorly utilized. The intrinsic factor may be absent, with the subsequent production of a macrocytic anæmia and a megaloblastic marrow, although why this type of anæmia is so uncommon is uncertain.

Anæmia is to be expected when absorption of any of the hæmopoietic factors is impaired. One cause of malabsorption may be intestinal hurry. Also, it is possible that alteration in the bacterial flora occurs following partial gastrectomy, which may alter the degree of absorption of some, if not all, of the various hæmopoietic factors.

Finally, patients following gastrectomy may alter their diet, which may eventually lead to the onset of anæmia.

A normochromic and normocytic anæmia was found in 14 patients, the majority of whom were women—a finding which is in agreement with a statement by Whitby and Britton (1953) that women are more prone to develop this type of anæmia. It is quite possible that this anæmia is due to some degree of protein deficiency in the diet.

Table III.—HYPOCHROMIC AND MICROCYTIC ANÆMIA

CASE NUMBER	HÆMOGLOBIN	RED CELLS	P.C.V.	M.C.V.	M.C.H.C.
	g. per 100 ml.	million per c.mm.	per cent	cu. μ.	per cent
34. G.U. and D.U.	7·9	4·6	34	74	23
35. D.U.	11·7	5·0	37	74	31
36. G.U.	7·2	3·9	29	75	25
37. G.U.	6·9	3·5	25	71	28
38. G.U.	8·5	4·7	31	66	28
39. G.U. and D.U.	6·3	4·4	28	63	23
40. G.U.	10·7	4·9	37	76	29
41. D.U.	8·2	4·3	32	75	26
42. G.U. and D.U.	11·3	5·0	38	76	30

G.U.—Gastric ulcer. D.U.—Duodenal ulcer.

It was expected that the number of women showing an iron-deficiency anæmia would be greatly in excess of the men, but this was not found. While these figures are too few in number for a statistical evaluation, they are of clinical significance.

No obvious difference in the blood-counts was found between patients operated on for gastric ulcer as compared with those for duodenal ulcer.

SUMMARY

The hæmatological pictures of 100 patients following partial gastrectomy are presented. In all cases the absolute values were determined. Anæmia was found in 44 cases. There were 14 cases of normochromic and normocytic anæmia, 19 cases of hypochromic and normocytic anæmia, 9 cases of hypochromic and microcytic anæmia, and 2 cases of macrocytic anæmia, 1 of the latter having a megaloblastic bone-marrow.

We are greatly indebted to Mr. H. T. Cox, for allowing us to carry out these investigations on his patients, and for his helpful criticism and advice. We would also like to thank Dr. Carolyn Rigby for assistance in some of the work.

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A CASE OF ACUTE PANCREATITIS AS A COMPLICATION OF THE PUERPERIUM

By ALAN DAVIES, London

In the puerperium the clinical picture of severe abdominal pain associated with pyrexia, tachycardia, nausea, and vomiting may be due to any of the acute abdominal emergencies, not directly related or attributable to the pregnant or puerperal state. There is, as well, the added hazard to diagnosis of a peritonitis consequent upon infection of the placental site. In the following report a case of acute pancreatitis occurring during the puerperium is described and the possibility that its aetiology is related to the puerperal state is discussed.

CASE REPORT

Mrs. H. H., aged 37 years, who had been delivered normally three weeks previously of a full-term live boy, was admitted on March 15, 1950, with severe abdominal pain. The pain had started suddenly 24 hours before admission. It was continuous and "vice-like", with periods of increased intensity. During the night prior to admission it had lessened slightly, but again became severe next morning. It was localized to the lower end of the sternum, radiating through to the back and was relieved to some extent by vomiting. Her bowels had not been opened for two days. During her pregnancy she had three episodes of vomiting associated with upper abdominal pain radiating through to the back. These occurred in the second, third, and fifth months of pregnancy. They had lasted a few days and had apparently been relieved by taking "salts and white powder". Apart from these she had been quite well, with no excessive vomiting or salivation.

In the immediate puerperium she had a bilateral calf vein thrombosis which had been treated by further rest in bed. Her lochia had been diminishing normally until the onset of this present attack, when it had increased in quantity and had become offensive.

On Examination.—She had a sallow complexion and her sclera were slightly jaundiced. Temperature was 99·8° F., pulse-rate 76 and respiratory-rate 24, with a blood-pressure of 130/90. Her tongue was furred and dry and she had fœtid breath. The abdomen was slightly distended and did not move on respiration. She was tender all over the abdomen, with rigidity in the upper quadrants. In both renal angles there was tenderness, more marked on the left than the right. No peristalsis was heard. The uterus felt bulky on rectal examination and both lateral pelvic walls were tender. There was a small para-umbilical hernia which was easily reducible. No other abnormal physical signs were found.

Investigations.—

Serum-amylase: 546 Smith Roe units per cent (normal range 30–140 units per cent).

Urinary diastase: 677 units (normal index up to 50).

White-cell count: 16,300/c.mm., with 91 per cent neutrophils.

Hæmoglobin: 90 per cent.

Straight radiograph of abdomen and chest: Considerable dilatation of the large intestine with gas, particularly in the region of the ascending colon. There was no free gas under the diaphragm, nor was there any evidence of opaque calculi in the region of the gall-bladder or the kidneys. No active disease of the lungs was seen.

Urine examination: No acetone, sugar, or albumin were present.

Cervical swab: A poor growth of anaerobes on culture.

A provisional diagnosis of acute pancreatitis was made, and this was confirmed by the raised serum-amylase level the following day. The patient was treated expectantly with intravenous glucose and saline, penicillin 500,000 units 4-hourly, streptomycin 1 g. b.d., and sulphatriad 2 g. 4-hourly.

Progress.—On March 16 (first day after admission) she still had severe, continuous abdominal pain. The pulse-rate had risen from 80 to over 90 and her temperature was 100·8° F. The abdomen was more distended, but there was return of peristalsis with passage of flatus. The lochia had not increased in quantity but there was some fresh blood contained in it.

On March 17 (second day) there was definite improvement in her general condition and, although the temperature had risen to 103·4° F., the pulse had started to settle and the abdomen was less tender. There was no evidence of subcutaneous staining (described by Grey Turner (1920) and Cullen (1918)) in the loin or around the umbilicus. This improvement was maintained and, by March 22 (sixth day), the patient was symptom-free, with no abnormal physical signs and eating a light diet.

The patient was discharged from hospital on March 31 (fifteenth day), 17 days after the onset of the pain. She was seen in the Out-patient Department two weeks after discharge, when there were no abnormal physical signs in the abdomen, but she did complain of some substernal pain which had been relieved by alkalis. She failed to attend for a cholecystogram, but was recently contacted and stated that she had been admitted to another hospital in June, 1950. A report from this hospital states that she had a further attack of abdominal pain and this was allowed to subside. She had a laparotomy in July, 1950, when a gall-bladder containing many small, soft stones was removed. The head of the pancreas was thickened and a biopsy showed appearances consistent with healed pancreatitis. The common duct showed no abnormality. Since her discharge from hospital she has been well, with no further recurrences of pain.

DISCUSSION

The earliest report of pancreatitis complicating pregnancy or the puerperium that I can find is that of Lawrence in 1838 and the latest review is that of Langmade and Edmondson (1951), who have collected 53 cases from the literature and added a further 9 of their own. They considered that some time limit after the termination of the pregnancy should be made if pancreatitis is to be related to the pregnancy or puerperal state. Six weeks post-partum was suggested as the maximum time. In 39 (73 per cent) it occurred during the puerperium, and in 28 of these there has been no previous symptoms referable to acute pancreatitis at any time before delivery. Eleven patients had a previous history of the disorder occurring during previous pregnancy, puerperal or non-puerperal state. The disease most commonly followed a first pregnancy.

The relationship between acute pancreatitis and pregnancy is obscure, but various aetiological factors have been suggested such as cholelithiasis, endocrine dysfunction, emboli, and changes in the pancreas following severe post-partum hæmorrhage.

In Langmade and Edmondson's review the gall-bladder was examined in 34 cases and calculi found in 12. In these cases the history and course of the disease differed in no way from those without calculi. Molander (1946) reported that cholelithiasis was present six times more frequently in cases of acute pancreatitis than in post-mortem examinations for other diseases. The exact relationship between gall-stones and pancreatitis is not definitely established, but as cholelithiasis is commoner in women one would expect that they would have a higher incidence of acute pancreatitis. In a small series of cases Molander found that the proportion of the disease in men and women was 10 males to 17 females. This, however, is not confirmed by other workers with longer series. Rich and Duff (1936) quote Gruber, who found in 94 post-mortems that the proportion of males to females was 58 to 36. Similar figures were given by Opie and Barker. Molander found that two-thirds of the male and one-third of the female cases had no gall-stones in association with their pancreatitis. Cholelithiasis is approximately twice as common in women as men, and Gross (1929) proves statistically that gall-stone is no commoner in parous women than in single women, or men. On this evidence it may be said that, although gall-stones may be present in cases of pancreatitis, they were not the cause of it, and may well have been formed by a biliary stasis due to some degree of obstruction to the duct by the fibrosed gland.

Two physiological disturbances have been suggested as aetiological factors in the development of acute pancreatitis in the non-pregnant state, and it is of considerable interest that both these factors can occur during pregnancy :—

1. There is an increase in the production of pancreatic trypsin and lipase. This was demonstrated by Picinelli and Villani (1940), quoted by Langmade and Edmondson (1951), using intraduodenal tubes to show that there is increased pancreatic activity during pregnancy. Presumably at this time there is increased vascularity and swelling of the pancreas. A comparison may be drawn with the salivary gland, which, histologically, is very much like the pancreas. Langmade and Edmondson point out that in pregnancy there may be excessive salivation associated with swollen, tender, salivary glands. Further evidence of a relationship is seen in epidemic parotitis in which pancreatitis may occur as a complication. In two post-mortems reported by Farnam (1922) the pancreas was found to be swollen in cases of mumps ; in neither case was pancreatitis a clinical complication.

2. Essential hyperlipæmia has been shown to be the cause of a case of relapsing pancreatitis. Klatskin and Gordon (1952) reported a case in which the serum-lipids and blood-cholesterol rose immediately prior to the onset of pain. In their opinion the pancreatitis was the result of the hyperlipæmia and not the cause of it. There is, in pregnancy, an increase in the blood level of fat and cholesterol. Taylor and Underhill (1925) showed that there is an increase in the fat content of the blood amounting to 50 per cent. This occurs from the third month and continues until the end of the first two weeks of the puerperium. There is also a corresponding increase in the cholesterol level.

It has been suggested that a vascular disturbance affecting the vessels of the pancreas may be important in the aetiology of pancreatitis ; this is applicable to the pregnant or puerperal state. Smyth (1940) reports that, in post-mortems on 40 cases of acute pancreatitis, thrombosis was frequently observed in the pancreatic vessels, occurring in 26 cases. He was able to reproduce the disease in localized areas by injecting mercury into the pancreatic artery of dogs. Whether the thrombosis that does occur is due to the action of the pancreatic enzymes on the vessels, or is the cause of the pancreatitis, is debatable.

This patient had, during her pregnancy, experienced symptoms which may have been due to acute pancreatitis in the second, third, and fifth months. She had not suffered from abdominal pain at any time before this, her previous pregnancies being uncomplicated. During this present pregnancy the patient had not suffered from excessive vomiting or salivation, her diet had been well balanced, and there had been no excesses of eating or drinking. She had shown evidence of a tendency to thrombosis and embolism in that she had had an attack of thrombophlebitis affecting the veins in her leg in the puerperium and, subsequently, had developed a further episode in the other leg.

In this present case and those of a series of 62 reported by Langmade and Edmondson (1951) in which it was possible to study the histories, no single factor was found to be common to all. In 13 cases of the 35 examined, including the present case, there were gall-stones found at operation or post mortem. In the discussion previously the conclusion was drawn, however, that there was no direct causal relationship between cholelithiases and pancreatitis, but biliary stasis due to pancreatic disease may lead to the formation of the stones. In the remaining cases there was no apparent causal factor. One may speculate that the complex electrolyte changes that occur both in pregnancy and the puerperium may in some way not yet appreciated precipitate the condition.

SUMMARY

A case is reported of a patient who developed an attack of acute pancreatitis three weeks after the delivery of a child. She had three similar but less severe attacks during pregnancy. At a later date she was found to have cholelithiasis. Two factors common to pregnancy and the aetiology of acute pancreatitis are discussed, with a comment referring to the present case.

Since this paper was written another paper has been published by Joske (1955) in which a further 6 cases of acute pancreatitis complicating the puerperium are discussed. Of these, 4 had gall-stones, but at laparotomy no obstruction could be demonstrated in the bile-duct or at the sphincter of Oddi. In these cases cholangiography showed a freely patent duct of Wirsung. It is suggested that the gall-stones are not primarily causing the pancreatitis, but are caused by it owing to the biliary stasis.

I should like to thank Mr. H. R. I. Wolfe for permission to publish this case.

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RENAL ECTOPIA IN SISTERS*

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THE incidence of renal ectopia as seen at autopsy lies between 1–600 and 1–1000 (Campbell, 1930; Thomas and Barton, 1936). Simple unilateral ectopia is the commonest form. The kidney may be low lumbar, iliac, or pelvic in position. Features which aid in differentiation from nephroptosis are multiple renal vessels arising at an abnormally low level, fixity of the kidney, and a short ureter. The case of a young woman recently investigated in this Unit acquires added interest from the fact that one of her sisters had suffered from a similar condition.

CASE REPORTS

I. J., an unmarried girl of 16 years of age, was referred by Dr. T. R. Bryant, of Tredegar, Monmouthshire. For nine months she had suffered from attacks of pain in the right iliac fossa. These were infrequent, lasted for a day or so, and were occasionally associated with nausea. She had lost a few pounds in weight, but there were no urinary or other symptoms.

On examination there was a discrete mass with a suggestion of lobulation deeply placed in the right iliac fossa and measuring some 8 × 3 cm. It lay in the line of the common iliac vessels and exhibited only very slight lateral mobility. It was not tender. Other systems were normal. Blood-pressure 130/70. An intravenous pyelogram showed a normal left pelvic outline. On the right side the renal pelvis was found to be overlying the L.5 and sacral vertebræ (*Fig.* 192). There was good function. The urine showed no abnormality on microscopy and culture.

Inquiry revealed that her sister G. M. J. had been admitted to Bristol Royal Infirmary in August, 1950, at the age of 21 years, under the care of Mr. Ashton Miller, who kindly allowed me access to her case records. For four years she had suffered from attacks of pain in the right iliac fossa and appendicectomy had been performed elsewhere with no relief of symptoms. On examination there was a slightly tender mobile mass in the right lower abdomen. Other systems were normal. Blood-pressure 130/70. An intravenous pyelogram confirmed the mass to be an ectopic kidney with incomplete rotation and kinking of the right ureter. The left kidney was normal in outline and function (*Fig.* 193). On Aug. 29, 1950, a right nephrectomy was performed and she has since remained well.

The parents are alive and well. There are five other sisters and two brothers, all of whom are in good health. Only one relative—a sister—consented to intravenous pyelography and in her case the renal anatomy was normal.

DISCUSSION

Laughlin (1942) reported a girl of 16 years with left unilateral renal ectopia. At the age of 20 (2½ years after nephrectomy) she delivered a full-term male stillbirth with incomplete rotation of the left kidney. The patient's sister, aged 17 years, was found to have congenital non-rotation of the right kidney, and her mother to have a right nephroptosis, "possibly congenital". No other record has been found of familial ectopia. Bilateral renal agenesis in siblings has been described on two occasions (Madisson, 1934; Schmidt, Hartley, and Bower, 1952). Two cases of congenital absence of one

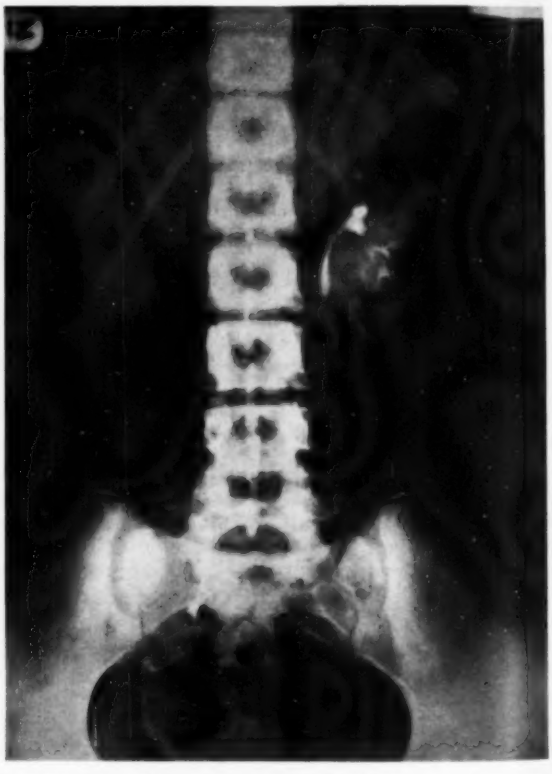

FIG. 192.—Intravenous pyelogram showing the right renal pelvis overlying the fifth lumbar and upper sacral vertebræ in the first patient (I. J.).

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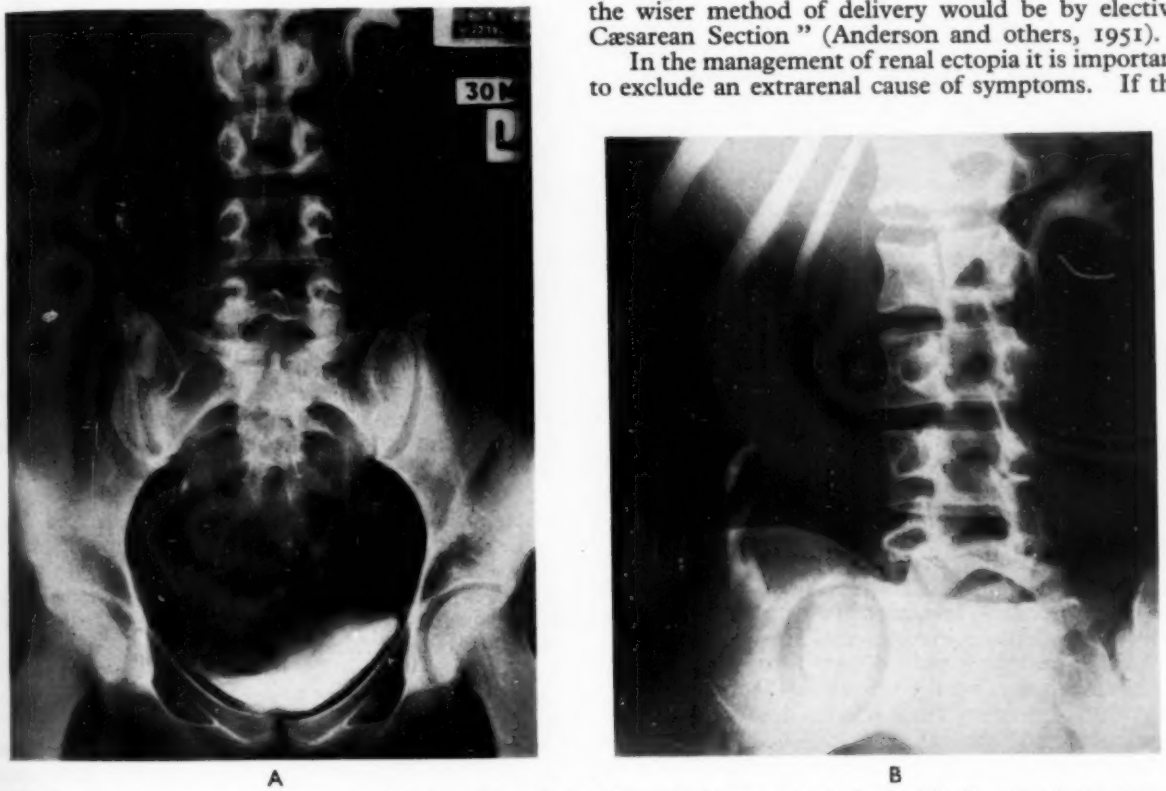
kidney in the same family were presented by Bound (1943). Polycystic disease of the kidney has a well-recognized familial incidence. The simultaneous presentation of identical twins with Wilms' tumours has been reported (Gaulin, 1951).

It may be that in a condition which is often clinically silent, and in which the diagnosis is evident only after special investigation or at autopsy, a small familial incidence may pass unobserved. The occurrence of renal ectopia in non-twin siblings suggests a hereditary factor rather than a random mutational defect or blighting factor during development. In an investigation of renal and genital defects in rats, Hain and Robertson (1937) concluded that such anomalies are inherited characters, though the mode of inheritance is not a straightforward one.



A B

FIG. 193.—A, Intravenous pyelogram showing the right renal pelvis incompletely rotated at the level of the iliac crest in the sister (G. M. J.); B, An oblique view.

While in both of the cases here described the renal ectopia was right-sided, reported cases of simple unilateral renal ectopia have usually, though not invariably, displayed a marked predilection for the left side (Anderson, Rice, and Harris, 1951; Daseler and Anson, 1943).

Renal ectopia is consistent with a healthy normal life span. In 27 autopsy cases, Campbell (1930) found that in none was the ectopia directly related to death. Five of the subjects were 50 years of age. Nevertheless ectopic kidney carries an increased liability to secondary renal diseases (by virtue of its incomplete development, abnormal location, and frequently imperfect drainage). Thompson and Pace (1937) found secondary pathological lesions in 22 of their 88 clinical cases of renal ectopia. There is a lowered resistance to infection which may be precipitated by instrumentation (Fetter and Smith, 1950). An ectopic kidney is more liable to injury than one in the normal position in the lumbar gutter, where it enjoys considerable protection (Huth, 1952). In women a pelvic ectopic kidney may complicate pregnancy and labour. With proper management, however, it is usually possible to effect a normal delivery, "but if all the renal tissue lies in the pelvis as in bilateral or solitary pelvic ectopia, the wiser method of delivery would be by elective Cæsarean Section" (Anderson and others, 1951).

In the management of renal ectopia it is important to exclude an extrarenal cause of symptoms. If the condition is silent or discomfort is not severe, and there is no secondary disease, radical treatment is not indicated.

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COMBINED INTESTINAL ASPIRATION AND FEEDING

A METHOD OF RESTING INJURED OR DISEASED SEGMENTS OF THE ALIMENTARY CANAL WITH PARTICULAR REFERENCE TO ANASTOMOSES AND FISTULÆ

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SINCE the days of John Hilton surgeons have recognized the importance of physiological and mechanical rest in natural healing processes. Further, orthopædic surgeons have realized that when disease is localized, normal function of adjacent healthy tissues should be encouraged.

But it is difficult to apply these principles to the surgery of the intestinal tract. Secretions cannot effectively be suppressed and their removal by continuous aspiration leads to problems of electrolyte imbalance and starvation.

The purpose of this paper is to describe a method conforming with these principles, whereby segments of the alimentary tract can be rested and yet functional continuity maintained.

This is achieved by means of a double tube lying in the lumen of the bowel. Through one of the two channels continuous suction is applied to the area to be rested, while the juices so aspirated are fed down the other channel together with liquid diet as required.

In this way negative pressure in the diseased segment is assured. Established fistulæ heal because they no longer leak, and potential fistulæ fail to form. Moreover, because there is no distension to encourage peristalsis, healing is promoted. Electrolyte balance is maintained because the secretions that are aspirated from the alimentary tract proximal to the diseased segment are fed back into the bowel distal to the lesion.

Jejunal feeding has proved useful not only in the treatment of fistulæ, but also after operations on the upper alimentary tract. In the latter cases glucose and water are absorbed within a few hours of operation and in about 48 hours when bowel sounds have returned the patient will absorb a 2000-calorie diet* and consequently suffer less of post-operative protein catabolism than usual (Beal, Cornell, and Gilder, 1954). With this treatment thirst, a common post-operative complaint with conventional treatment, is conspicuous by its absence.

During the last six years this method has been used in this hospital in the treatment of injuries and fistulæ of the œsophagus or duodenum, and it has in the last two years been applied to the care of patients after operations such as partial gastrectomy and œsophago-gastrectomy which involve anastomoses in the upper alimentary tract.

A double tube was first described by Williams and Walsh (1930). They introduced it through a gastrotomy in cases of perforated duodenal ulcer with stenosis. The outer rubber tube drained the stomach and the inner tube, a ureteric catheter, fed fluid through the stenosis into the duodenum.

The use of two tubes, one for suction and one for feeding, was suggested by Perl (1935) as a treatment for duodenal fistula, and Abbott and Rawson (1939) described a double-lumen tube (size 16F.) for gastric aspiration and jejunal feeding after gastro-enterostomy; its use in maintaining nutrition after gastric operations was shown by Stengel and Ravdin (1939).

It is, however, only recently with the advent of plastic tubes that this method has become easy to apply.

PREPARATION AND INTRODUCTION OF THE TUBES

The first step is to pass a 'polythene' tube with a bore of 1·6 mm. and an external diameter of 2·6 mm. through the injured area into the healthy bowel beyond.

In patients undergoing operation this is done by the combined manipulations of surgeon and anæsthetist; it is important that the advancing end of the tube should be guarded by a rubber tip.

In other patients the 'polythene' feeding tube is prepared by marking it with thread at three points which when at the nose will indicate positions of the end of the tube at cardia, pylorus, and duodenum (45 cm., 60 cm., and 75 cm. from the tip). A mercury-laden finger-stall is attached to the end of the tube so that its (onward) passage can be assisted by gravity and peristalsis. This tube is then passed through the nose and encouraged on its travels down the alimentary tract by positioning the patient in the following way:—

1. The tube is passed into the stomach, and the patient is sat bolt upright for an hour or so to get the bag into the pyloric antrum. In patients with abdominal distension this may be difficult because the stomach is folded up against the left diaphragm (*Fig.* 194) and the tube tends to be deflected off the incisura and curl back up into the fundus.

2. When the tube has reached the pyloric antrum the patient is laid on the right side for a few hours with the third mark on the tube (75 cm.) at the nose. A radiograph is then taken to show its position; and it is helpful to outline the tube by injecting a little barium down it just before this plate is exposed.

3. Once the tube arrives in the duodenum, more of it is passed into the nose, and the patient is laid on the *left* side to carry the bag over the vertebral column, which in the supine position represents quite a steep climb in the third part of the duodenum. Hereafter passage down the small bowel is rapid. When the tube is well past the diseased or injured area it is marked with a thread ligature at its point of emergence from the nose, and the second phase begins.

* Radio-active iron is even absorbed at this stage (Smith, 1955).

A larger plastic tube (polyvinyl chloride ('P.V.C.'), 'Portex' for instance) of 5-mm. internal diameter (*see* Appendix, 1), with side holes cut at its distal end, is lubricated internally with paraffin and threaded over the long length of 'polythene' tube emerging from the patient's nose. Then, after external lubrication with anæsthetic ointment, it is pushed down by the 'railroad' method, over the 'polythene' tube which is kept stationary in the nose. The side holes in the 'P.V.C.' tube are marked with radio-opaque metal clips or wire to facilitate the positioning of the tube under X-ray control. Once

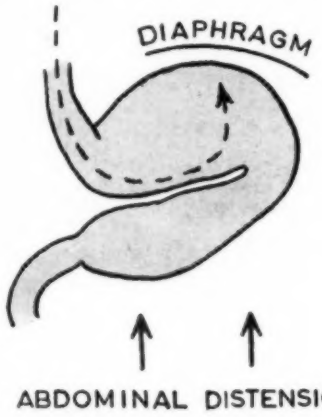

FIG. 194.—Intestinal distension causes the stomach to be folded up against the diaphragm. The exaggerated incisura is a barrier to the passage of an intestinal tube.

the tube is in position without slack it is fixed by plastic adhesive tape to the nose.

To enable aspiration to be done through the 'P.V.C.' suction tube, a short segment of larger bore tube of the same material is taken, and the end of the 'polythene' tube is introduced into this and brought out through its wall by means of a broad-gauge hollow needle. This broader tube is now slid down the 'polythene' and over the first 'P.V.C.' tube as a sleeve to make an airtight joint (*Fig.* 195).

MANAGEMENT OF THE DOUBLE TUBE

Either intermittent or continuous suction is applied to the suction tube according to the condition being treated. For continuous aspiration a negative pressure of 6 to 12 cm. of mercury has proved satisfactory. Kinking and collapse of the suction tube is impossible because its walls are held apart by the inner feeding tube.

The aspirate is filtered and is fed back down the feeding tube together with a liquid diet. This diet varies from 5 per cent glucose or electrolyte solutions in the immediate post-operative period to a liquid diet supplying approximately 3000 calories per day when any initial ileus has relented (usually after 36–48 hours). In addition to this fluids are allowed by mouth, except to patients with œsophageal lesions.

In the rare cases where paralytic ileus develops, jejunal feeding is discontinued and fluids are given intravenously. To decompress the distended bowel the suction tube is 'railroaded' down the feeding tube into the jejunum to act like the conventional Miller-Abbott or Cantor intestinal tube.

In patients with an external fistula suction should never be applied to the external orifice, because it increases the discharge and prevents healing.

APPLICATION OF THE METHOD WITH ILLUSTRATIVE CASES

As a Post-operative Treatment

1. Œsophagectomy or Total Gastrectomy.—The anastomoses in these operations, particularly if œsophagogastric, are not always reliable. The blood-supply to both œsophagus and stomach is often

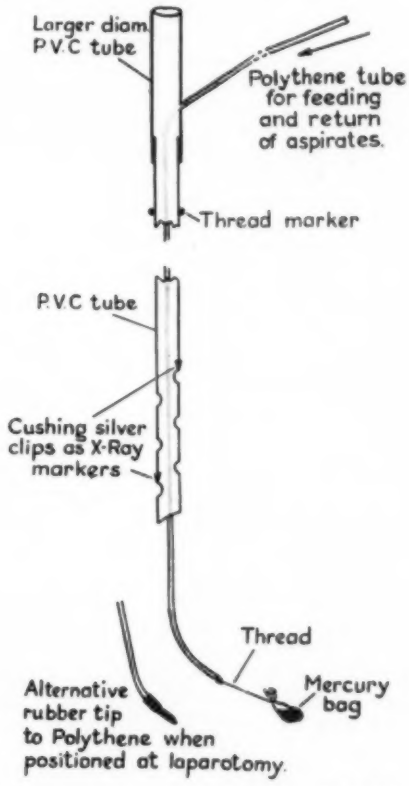
Larger diam P.V.C tube
Polythene tube for feeding and return of aspirates.
Thread marker
P.V.C tube
Cushing silver clips as X-Ray markers
Thread
Mercury bag
Alternative rubber tip to Polythene when positioned at laparotomy.

FIG. 195.—Diagram of the feeding and suction tubes. The polythene feeding tube enters the side wall of a short segment of 'P.V.C.' tube that fits like a sleeve over the end of the main suction tube. Both tubes are marked with thread in case adjustment is necessary.

meagre, the latter particularly so if the gastro-duodenal artery is arteriosclerotic. Moreover, in the post-operative period alimentation is usually restricted to the intravenous route for upwards of a week—a period of malnutrition that undermines the patient's strength and does not favour healing at the anastomosis.

These considerations, which lead some surgeons to perform a jejunostomy, prompt us to pass a feeding tube into the duodenum and to keep the anastomosis at rest by continuous suction (*Fig.* 196). This method has been used in several such operations with satisfactory results and patients have survived easily when with more conventional treatment a fatal result would have been expected.

The tubes are passed at operation through the anastomosis after completion of its posterior layer; the end of the feeding tube is passed into the

duodenum or jejunum and the tip of the suction tube lies about 2 in. beyond the anastomosis.

Until the eighth post-operative day no fluids are allowed by mouth, and continuous suction is applied to the outer tube. In the average case feeding through the inner tube is started after 12 hours, and 4 pints of 5 per cent glucose are given in the next 24 hours. This is then changed to milk, and in the absence of abdominal distension high-calorie feeding is begun on the third day. All aspirate is filtered and re-introduced down the feeding tube.

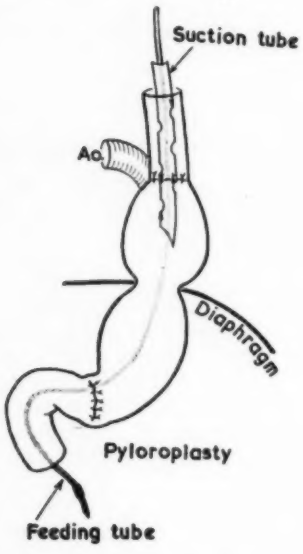

FIG. 196.—Shows the arrangement of tubes after œsophagectomy and œsophago-gastrostomy.

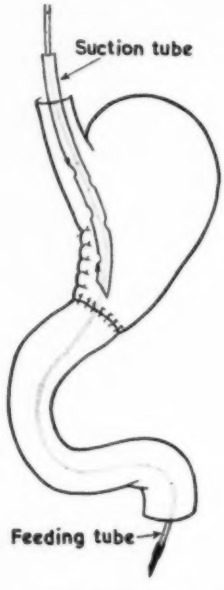

FIG. 197.—Management of suction and feeding tubes after Billroth I gastrectomy.

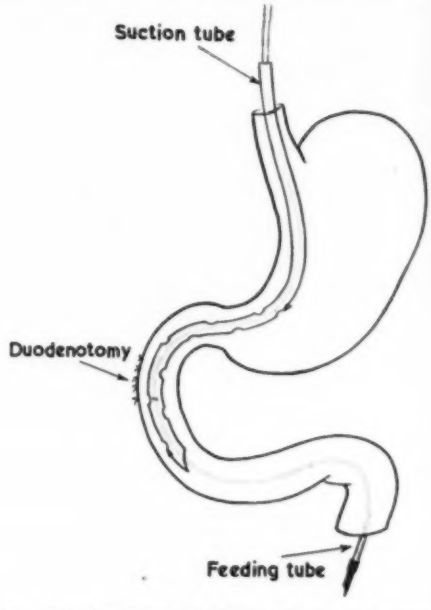

FIG. 198.—Arrangement of tubes in the post-operative management of operations on the duodenum.

2. After Partial Gastrectomy.—The conventional treatment by stomach aspiration and intravenous, intramuscular, or rectal alimentation usually gives good results; but on occasions, particularly after the Billroth I operation, it has to be prolonged for a week or more. All these methods of administering fluids are unpleasant for the patient, and prone to cause complications such as thrombophlebitis.

The following routine eliminates these troubles and greatly simplifies the after-care :—

Before the patient comes to the theatre, a ' P.V.C.' stomach tube with a bore of 5 mm. is passed through the nose. When the surgeon has completed the posterior layer of the anastomosis he passes a chemically sterilized ' polythene ' tube up the lumen of the wider tube, and it is then grasped by the anæsthetist; the other end of the ' polythene ', guarded by a rubber tip, is manipulated down through the duodenum and about 3 in. into the jejunum. The operation is completed in the usual way (*Fig.* 197).

The feeding programme and re-introduction of aspirate is the same as after œsophagectomy, but clear fluids are allowed by mouth. In the ordinary case where the surgeon has confidence in the anastomosis, aspiration is intermittent so that stomal function can be assessed. When the stomach is emptying satisfactorily the suction tube is removed, but the feeding tube is left in situ for a further 12 hours in case suction has to be re-instituted.

Up to the present more than 150 patients have been treated in this way after gastrectomy. Electrolyte balance is no longer a problem, because all aspirate is re-introduced into the jejunum; the patient is better nourished—particularly important in the poor-risk case—and both patients and house surgeons benefit from sleep undisturbed by temperamental intravenous infusions.

3. After Pancreatico-duodenectomy.—One of the main dangers of this operation is leakage at the pancreatico-jejunal anastomosis, and so the use of the two-tube method should substantially decrease the risk of this operation.

4. After Operations on the Duodenum.—Some operations on the duodenum such as trans-duodenal sphincterotomy of the ampulla of Vater and choledocho-duodenostomy when treated post-operatively in the customary way are frequently followed by duodenal fistula. This dangerous complication has not arisen since it has been the practice to put the operation site at rest with continuous suction by the method described.

Fig. 198 shows the appropriate arrangement of the tubes, which should be kept in situ for eight days.

Use of the Method in the Upper Alimentary Tract as a Primary Treatment of Fistulæ and Injuries

Injuries of the alimentary tract are unlikely to lead to perforations or fistulæ, and established fistulæ are likely to heal if the affected segment of gut is kept empty and at rest. Described below is a series of cases illustrating the use of the method in lesions of the pharynx, œsophagus, and duodenum; an extension of the method of lesions to stomach and small intestine is discussed.

1. Pharyngeal Fistula.—One such case has been treated by this technique; but success was only partial because the fistula traversed the thorax and respiratory movements caused a continuous ebb and flow of mucus which prevented healing.

Suction proved effective in removing saliva and mouthwashes, some of which would otherwise have entered the fistula and hampered healing. Since the continuous aspiration of air from the pharynx could not be avoided, a pump with a high capacity (Sprengel water pump) proved necessary to maintain sufficient negative pressure to suck up the viscid sputum.

Case P. W., R.I. No. 182176.—

A woman of 44 was admitted to the Thoracic Surgical Unit. Four days previously gastroscopy had been unsuccessfully attempted at another hospital—it had resulted in a ½-in. perforation of the posterior pharyngeal wall at the classical site.

Treatment was at first confined to transpleural drainage of the resultant mediastinal abscess and feeding by gastric tube. The perforation failed to heal and swallowed fluids oozed from the intercostal drain.

Nineteen days after the injury external pharyngotomy was done, and the perforation was sutured. The incision healed, but an intractable staphylococcal infection of lung and pleural space developed and the pharyngo-mediastinal fistula reappeared.

Thirty-two days after the injury a test feed of 12 oz. resulted in a discharge of 9 oz. from the intercostal drain.

Pharyngeal suction was now begun; *Fig.* 199 shows the arrangement of the tubes and *Fig.* 200 the effect on the volume of discharge from the fistula. Suction was maintained for 51 days. The fistula became smaller but did not heal. This failure was probably due to the ebb and flow of mucus that was observed in the pharyngeal end of the fistula with each respiration. After a further attempt at surgical closure of the perforation a small cervical fistula developed and the mediastinal track healed.

It was not until a year after her injury that the patient began to swallow normally.

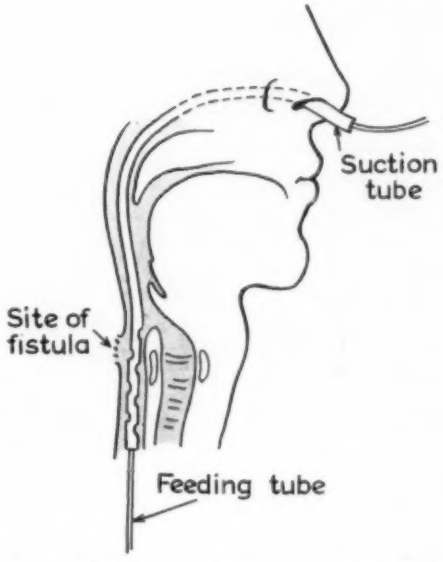

FIG. 199.—Arrangement of tubes in a case of pharyngeal fistula.

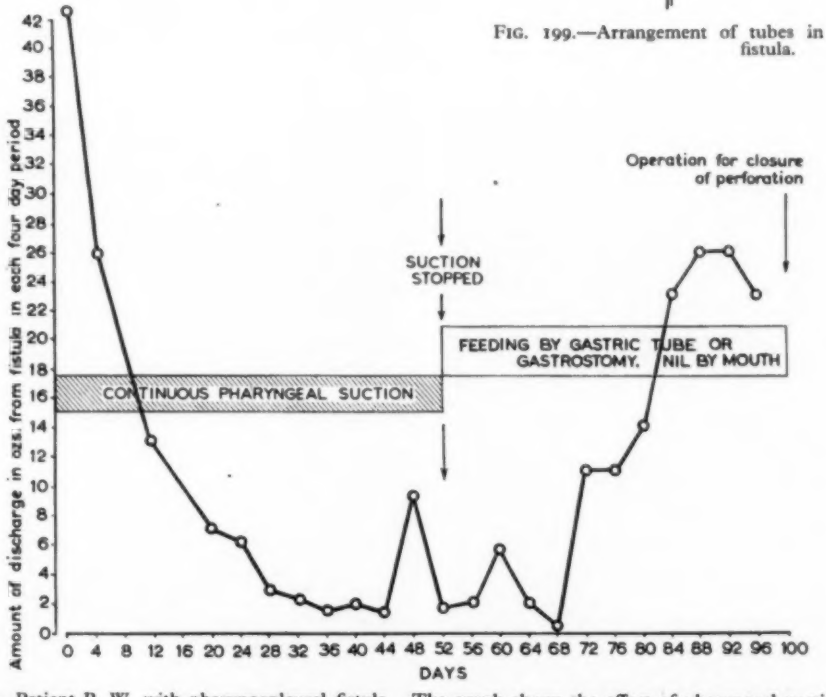
Amount of discharge in ozs. from fistula in each four day period
0 2 4 6 8 10 12 14 16 18 20 22 24 26 28 30 32 34 36 38 40 42
Operation for closure of perforation
SUCTION STOPPED
FEEDING BY GASTRIC TUBE OR GASTROSTOMY. NIL BY MOUTH
CONTINUOUS PHARYNGEAL SUCTION
0 4 8 12 16 20 24 28 32 36 40 44 48 52 56 60 64 68 72 76 80 84 88 92 96 100
DAYS

FIG. 200.—Patient P. W. with pharyngopleural fistula. The graph shows the effect of pharyngeal suction on the amount of discharge. After the two-tube treatment had been abandoned the volume of discharge increased.

Comment.—It seems that in this condition pharyngeal suction, although helpful, is not curative; and that it is not to be used to the exclusion of conventional treatment such as pharyngotomy or pharyngostomy.

2. Œsophageal Injuries.—The two-tube method is of use as a primary method of treatment when direct surgical repair is contra-indicated, and particularly in lacerations the depth of which is not known. It is also useful as a secondary treatment after surgical repair of these lesions.

So long as negative pressure is maintained in the lumen of the œsophagus escape of œsophageal contents into the mediastinum is impossible ; and under these circumstances antibiotics are likely to prevent the development of mediastinitis.

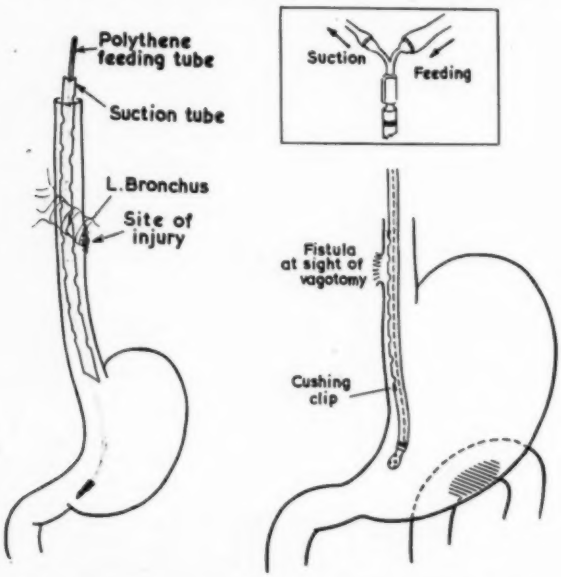

FIG. 201.—Shows the arrangement of tubes in the treatment of œsophageal injuries.

FIG. 202.—Scheme of treatment of an œsophageal fistula by means of a modified Miller-Abbott tube.

Fig. 201 shows the usual scheme of treatment of these patients ; though in the following case, which was treated prophylactically, the feeding arrangement was modified because a gastrotomy had to be done to remove the foreign body, and consequently the feeding tube was introduced through this opening.

Case E. B., aged 38, an inmate of H.M. Prison, R.I. No. 44127.

Some years previously Sir Hugh Cairns had performed a prefrontal leucotomy on this man in an attempt to cure his delinquency. On this occasion he was admitted from H.M. Prison having swallowed a naked Ever Ready safety-razor blade.

Radiographs showed the blade to be in the stomach. At œsophagoscopy 4 hours later cuts were seen below the cricopharyngeus and at the level of the left bronchus, but it was difficult to gauge their depth.

The razor blade was removed from the stomach by gastrotomy, so in this case the feeding tube was introduced through this opening across the stomach into the duodenum. A plastic suction tube with multiple side-holes was put down into the œsophagus with its tip projecting into the stomach. All the aspirate was re-introduced into the duodenum together with his feeds. On the eighth day the tubes were removed and he began to swallow normally.

Comment.—In this case no mediastinitis developed, but the cuts may not have penetrated the whole thickness of the œsophageal wall.

3. Œsophageal Fistulæ.—Conventional conservative treatments of this condition are often successful, but it seems likely that the two-tube method promotes more rapid and certain healing.

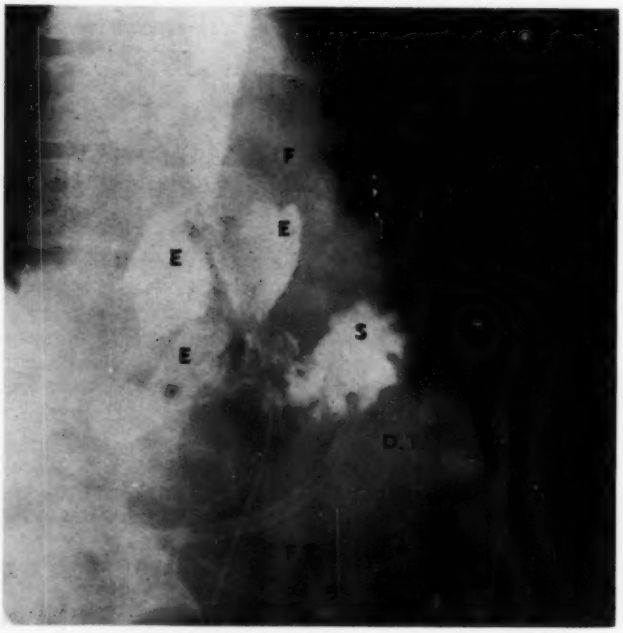

FIG. 203.—Œsophageal fistula after vagotomy. Barium study at beginning of treatment. Showing extensive extravasation (E) of barium from the œsophageal fistula (F). S, Stomach ; D.T., Abdominal drainage-tube ; F.T., 'Polythene' feeding tube in duodenal loop.

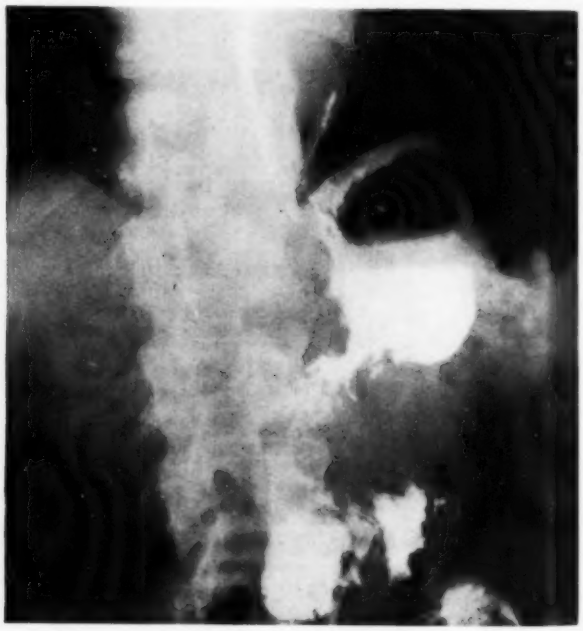

FIG. 204.—Œsophageal fistula healed after the two-tube treatment. Barium has been trapped in the tissues at the site of the fistula (F).

Case J. I., aged 77, R.I. No. 153251/52.—

Underwent a vagotomy and gastro-enterostomy for duodenal ulcer that had caused much pain and two hæmatemeses. During the vagotomy, which was difficult because of adhesions, a hole was inadvertently made in the œsophagus. This was repaired with catgut. Post-operatively the patient was treated by intermittent gastric aspiration without any feeding by the mouth apart from 12 oz. of fluid given in error on the 1st day. On the

3rd day a fistula developed through his abdominal wound and discharged from ½ to 9 oz. of fluid per day.

On the 16th day, after the major part of a test feed had been seen to emerge from the fistula, the two-tube treatment was begun. Thereafter drainage from the fistula was not measurable. Intravenous fluid therapy was now discontinued, and the patient's general condition improved *pari passu* with his improved nutrition. After 8 days of this treatment the fistula was well healed and he began to eat normally.

At the time when this patient was treated suitable plastic tubes were not available, so a Miller-Abbott tube was modified for the purpose (*Fig.* 202). Although successful in this case, such a tube is not ideal because its small bore makes it prone to blockage.

The following similar case has been treated with plastic tubes :—

Case J. O., aged 57, R.I. No. 168242/52.—

A very cachectic man weighing under 7 st., with stenosis of the stoma of a Billroth I gastrectomy. During the vagotomy œsophageal laceration occurred, as in *Case* J. I. described above. It was repaired and the two-tube method was instituted as a prophylactic measure. In spite of this a fistula appeared, but it was found that in error strong suction had been applied to the abdominal drainage-tube, and it seemed that the resulting pressure gradient across the suture line had caused it to leak. *Fig.* 203 is a radiograph showing this fistula. Suction to the drainage-tube was stopped and after 14 days of the two-tube treatment the fistula was healed (*Fig.* 204).

Comment.—There is little doubt that the two-tube treatment saved the lives of both these patients. The second case, J. O., illustrates the danger of applying suction direct to the wound drainage-tube.

4. Gastric Fistulæ.—The conservative treatment of perforated peptic ulcers by continuous gastric suction is well established. Consequently it seems likely that gastric fistulæ would respond equally well to this type of treatment. In the one such case encountered the fistula arose from the anastomosis of a Billroth I gastrectomy and attempts to introduce the two tubes were unsuccessful.

Case R. D., male, aged 39, R.I. No. 125212.—

This man underwent a Billroth I gastrectomy for a posterior duodenal ulcer. No feeding tube had been put into the small bowel at operation, and when a fistula appeared on the 4th day it proved impossible to persuade the polythene tube to go through the anastomosis, in spite of a leader consisting of a thread loaded variously with a mercury-filled bag or lead pellet. As a result of the continued local sepsis the patient developed a portal-vein thrombosis. His ascites eventually disappeared after a long course of anticoagulants.

The difficulty of intubating the Billroth I anastomosis has previously been encountered and is a further argument for the adoption of the two-tube method in the post-operative treatment of these cases.

5. Duodenal Fistulæ.—This condition shows a fatality-rate of about 50 per cent when treated by conventional methods, and the larger the fistula the more lethal it is. The author reviewed this condition (Gardner, 1951) and described the use of a modified Miller-Abbott tube in its treatment. The Miller-Abbott balloon was used as an internal obturator while nourishment was provided through the channel that is normally used for suction. The two-tube method of treatment described in this paper appears to be equally efficacious and technically much simpler to apply.

Case S. P., aged 71, R.I. No. 173884.—

A jaundiced old man underwent a cholecystectomy for acute cholecystitis and transduodenal choledocholithotomy for a stone impacted in the ampulla of Vater. On the 15th post-operative day he developed a fistula from the site of the duodenotomy which discharged about 30 oz. (900 ml.) per 24 hours. Under the two-tube treatment the fistula ceased to discharge and healed in 6 days. The patient made an excellent recovery. *Fig.* 198 shows the arrangement of the tubes and *Fig.* 205 is a photograph of the patient undergoing the treatment.

FIG. 205.—Photograph of a patient undergoing the two-tube treatment for duodenal fistula.

Case A. C., aged 61, R.I. No. 74513/47.—

Twenty-four years previously this patient had undergone two operations for a stricture of the common bile duct. Recently she had suffered recurrent jaundice and fever. At operation stones were found above a stricture in the common bile-duct ; the stones were removed and the duct was transected above the stricture. The dilated open end was then anastomosed to the uppermost part of the duodenum. Unfortunately a duodenal fistula developed from this anastomosis, which drained 128 oz. on the 7th day, and 85 oz. on the 8th day. The two-tube method was put into operation on the 9th day (*Fig.* 206), and thereafter the daily drainage was 20, 24, 40 (with the tube intermittently blocked), 12½, 5½, 1. On the 14th day the fistula healed and the patient recovered. The third case was one of a terminal duodenal fistula after Polya gastrectomy. Here the two-tube method was used to overcome a complete functional block at the outlet of the stomach.

Case F. P., aged 46, R.I. No. 57910.—

Underwent a Polya gastrectomy for duodenal ulcer, the pyloric antrum being closed by the Bancroft

technique. After the operation the anastomotic stoma obstinately refused to function, and during the first two weeks the total volume of stomach aspirate exceeded the oral intake by 401 oz. (12 l.).

On the 14th day after the operation an abscess lying between the anastomosis and the liver, which had arisen from a leak in the pyloric stump, was drained, and a stoma was done. In spite of this the aspirations in the following week exceeded the intake by an average of 72 oz. (2160 ml.) per day. It was now obvious that the obstruction was not mechanical, but functional, and it seemed possible that the high-protein jejunal feeds were in some way inhibiting the function of the bowel above the site of their entry. This in fact proved to be the case, for when on the 50th post-operative day these feeds were discontinued, the stoma immediately began to function normally.

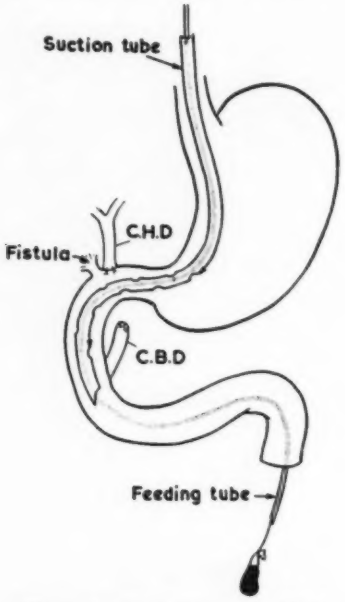

FIG. 206.—Shows the arrangement of tubes in the treatment of a duodenal fistula following choledochoduodenostomy.

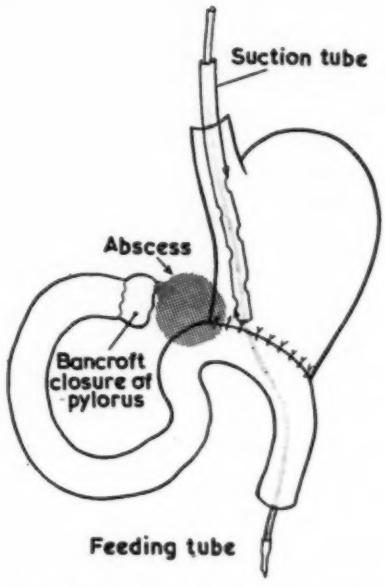

FIG. 207.—Shows the arrangement of tubes in the treatment of a functional obstruction at the anastomosis of a Polya gastrectomy.

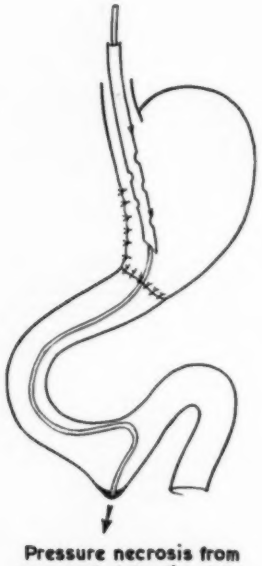

FIG. 208.—Showing how the bowel wall can be perforated by the unguarded tip of a polythene tube.

double tube was passed through the gastrojejunal anastomosis (*Fig.* 207).

During the next four weeks the stoma still did not function, but the patient was nourished via the jejunal tube and kept in electrolyte balance by returning all the gastric aspirate into the jejunum. On the 42nd post-operative day, after barium had repeatedly failed to pass the stoma, a Finney-type plastic enlargement of the

Continued leakage from the pyloric stump made two further operations for subhepatic abscesses necessary, but the patient eventually recovered and was discharged from hospital six months after his original operation.

Comment.—Doubtless the failure of the gastro-jejunal anastomosis to function was at first the result of the subhepatic abscess. Later on, however, it appeared that the jejunal feeds were inhibiting the function of the small bowel above the point of entry. If this was so it may throw light on the dysfunction after œsophago-jejunostomy of some intestinal loops of the Roux 'en Y' type and possibly also on the syndrome that may follow a (Polya) gastrectomy. This subject is undergoing further investigation.

6. Small-bowel Fistulæ.—In the absence of obstruction such fistulæ usually close spontaneously. The two-tube method is, however, applicable, and might be of use in special cases.

EVALUATION OF THE TREATMENT

Several very frail patients have survived total gastrectomy or œsophagectomy with the two-tube treatment. These recoveries would have been improbable with conventional treatment and are attributable to the maintenance of nutrition and electrolyte balance with this technique.

In the same way five of the patients with fistulæ—viz., J. O., J. I., S. P., A. C., and F. P.—whose case histories are given above, were unlikely otherwise to have recovered.

After operations on the upper alimentary tract the method has proved most useful :—

1. Intravenous infusions and their complications are avoided.

2. Early ambulation is made easier since the patient is free to move his limbs.

3. Problems of electrolyte balance and nutrition disappear because all aspirates are returned into the small bowel together with the liquid diet.

4. If paralytic ileus supervenes the suction tube can easily be fed down into the small bowel by the 'railroad' method to decompress it.

5. If enterocolitis occurs the appropriate antibiotic can be introduced down the jejunal feeding tube to a point near the watershed of the fluid coming up and the fluid going down the gut.

6. In uræmic and comatose patients the two-tube method is probably the safest way of feeding, as it prevents inhalation of vomit. The stomach is kept empty and small feeds of high caloric value are given directly into the small bowel.

No troubles have arisen from the presence of tubes passed through œsophageal, gastric, or intestinal anastomoses. The tube in the nose is uncomfortable, but no worse than that of the conventional transnasal stomach tube.

The only mishap in this series of more than 100 patients was a perforation of the third part of the duodenum :—

Case E. B., aged 75, R.I. No. 201727.—

This patient was a very sick man; he had had a hæmatemesis from a duodenal ulcer and died 5 days after gastrectomy from subtotal consolidation of his lungs and jaundice. The duodenal perforation was an incidental finding at autopsy; it had been caused by the unguarded tip of the polythene tube, which had curled up in the third part of the duodenum and caused pressure necrosis of the wall (*Fig.* 208). This danger is now avoided by using a soft rubber tip to the tube, and making sure at operation that this is lying free in the upper jejunum.

As with all treatments of this type, the two-tube method requires attention to detail if it is to be successful. In particular the tubes must not be allowed to block. In the treatment of fistulæ suction must be continuous and applied only to the suction tube. Suction applied to the external opening of the fistula encourages leakage and prevents healing.

SUMMARY

1. In the surgery of the alimentary tract the principle of resting the diseased part and maintaining function in the adjacent healthy regions has hitherto been neglected.

2. A technique is described which uses for this purpose concentrically arranged plastic feeding and suction tubes after operations and injuries of the alimentary tract and in the treatment of fistulæ.

3. Advantages of the method are that intravenous infusions are avoided, electrolyte balance is no longer a problem because all aspirates are returned to the alimentary canal, and nutrition is maintained by jejunal feeding.

4. In one patient the feeding of a protein-rich diet into the jejunum inhibited the bowel proximal to its site of entry. This may throw some light on the post-Polya gastrectomy syndrome and on the dysfunction after œsophago-jejunostomy of some intestinal loops of the Roux 'en Y' type.

5. Uses and modifications of the technique in paralytic ileus, enterocolitis, and uræmia are discussed.

I am much indebted to Mr. Elliot-Smith, Mr. Moloney, Mr. Corry, and Mr. Pile for encouragement and permission to publish details of their cases; and to Miss McLarty and Mr. Tugwell for the diagrams and photographs.

APPENDIX

1. Types of Tube.—For suction a P.V.C. tube of 5-mm. bore and 7-mm. diameter ('Portex' 10) has been found most satisfactory; larger diameter tubes tend to flatten with suction. Some batches of P.V.C. tubes are softer than others; if too soft they may be hardened by immersion in ether for 2–4 hours.

For feeding polythene tube size 3, 1·6-mm. bore is satisfactory. A smaller size can be used, but is more liable to block.

2. Suggested Diet for Jejunal Feeding.—

40 oz. Whole Milk
4 Eggs
6 oz. Full Cream Dried Milk (Cow & Gate)
4 oz. Glucose
2 oz. Soyolk
2·5 g. Sodium Chloride
3 gr. Ferrous Sulphate
3 drops Radiostoleum
2 tsps. Befortis
200 mg. Ascorbic Acid.

Calories 2754

This mixture is diluted with milk or water to give the appropriate fluid intake for 24 hours. If it proves too thick the Soyolk can be omitted and the glucose increased.

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TRAUMATIC ARTERIAL SPASM
ITS RELIEF IN MAN AND IN MONKEYS

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THE evidence from previous work (Kinmonth, Simeone, and Perlow, 1949; Kinmonth and Simeone, 1952; Kinmonth, 1952) indicates that traumatic arterial spasm is myogenic in nature and independent of nervous control. It can be relieved by applying smooth-muscle relaxing drugs such as papaverine directly to the vessel. Most of the previous work was done using the rabbit as the experimental animal. It has since been possible to extend the laboratory studies to primates and also to gain experience in the use of the method in clinical surgery. The results are reported here.

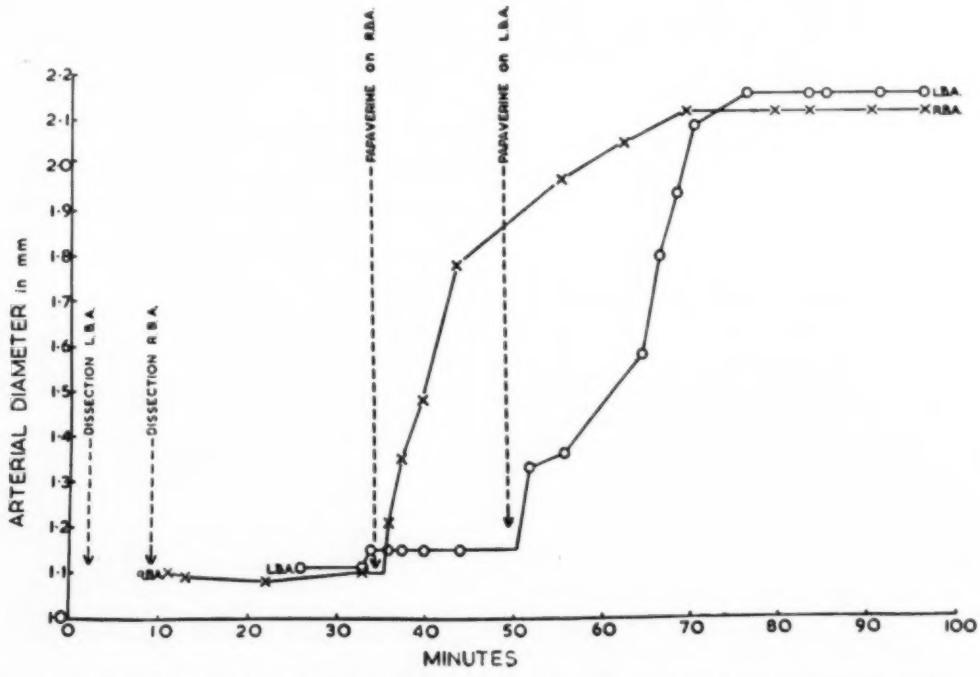

FIG. 209.—Traumatic spasm of brachial arteries of rhesus monkey relieved by 2·5 per cent papaverine sulphate.

ARTERIAL SPASM IN MONKEYS

The animals studied belonged to the family of old-world monkeys (cercopithecidæ) and the individual species were sooty mangabeys (*Cercocebus torquatus*), monas (*Cercopithecus mona*), and *rhesus* (*Macaca mulatta*). No species difference was found in the behaviour of the vessels or in their response to treatment.

Method.—The animals were anæsthetized with intravenous pentothal 50 mg./kg. followed by endotracheal oxygen supplemented when necessary with endotracheal oxygen, nitrous oxide, and trilene. In the latter half of the series intraperitoneal nembutal, 30 mg./kg., was used. The type of anæsthesia did not affect the response of the vessels. The skin was incised over the femoral artery just below the groin or over the brachial artery on the inner side of the arm and the vessel dissected free. The knife was used first, then blunt dissection. This was often sufficient to make the vessel contract into a white cord with closed lumen. If it did not do so rubbing with dry gauze or stretching was resorted to. On most occasions full spasm was achieved in this way, but sometimes the vessel did not react or only partly contracted and that vessel was then left and another tried or the experiment abandoned. No explanation could be found for this variation in irritability. It bore no relation to species, sex, anatomical site, or individual animal. Such variation has also been seen in the rabbit. When spasm was fully established a thread was placed under the vessel to mark a constant level at which its diameter was measured at intervals through a dissecting microscope with a micrometer eyepiece. The same things were done with the artery on the opposite limb as far as possible at the same time. The exposed vessels were kept covered or moist with warm Ringer-Locke solution.

Table I.—INCIDENCE OF SPASM IN MONKEYS

		Brachial	*Femoral*
Arteries observed	30	13	17
Spasm: Full	16	6	10
Partial	8	2	6
None	6	5	1

When it was desired to apply papaverine solution the Ringer-Locke was first gently sucked off with a pipette and the papaverine (2½ per cent papaverine sulphate) afterwards dropped on to the vessel with another pipette; sufficient papaverine solution was used to keep the vessel covered if possible.

Results.—Ten monkeys were used and experiments were completed on thirty arteries, anæsthetic complications or hæmorrhage interfering with observation in the others. The incidence of spasm is shown in *Table I*. Full spasm, as described above, occurred in 16 vessels and partial in 8. In 6 vessels spasm could not be induced.

A solution of papaverine sulphate was applied to the spastic vessels. Nine cases of full spasm and 3 of partial spasm were thus treated and in every case relaxation occurred. The dilatation usually became noticeable to the naked eye some 5–15 minutes after application. Observation through the microscope showed that there was a short latent period of ½–3 minutes, followed by a sharp increase in diameter and an alteration in the colour of the vessel wall from white to pink. The course of events is well shown in *Fig.* 209.

The behaviour of 6 fully spastic vessels untreated by papaverine is shown in *Fig.* 210 and contrasted with that in 6 other vessels where papaverine was used. In no instance did such abrupt or rapid dilatation occur in any of the untreated vessels, although it was a constant feature after application of papaverine.

The appearances of a spastic femoral artery in a female sooty mangabey monkey before and 20 minutes after applying 2·5 per cent papaverine are shown in *Fig.* 211. Some slight brown discoloration of loose blood-clot present in the wound was often seen, but it appeared to be quite harmless; for the wounds all healed well and the vessels, as judged by palpation of pulses, remained patent after dilatation.

The results obtained in these experiments in the monkeys showed that papaverine consistently relaxed traumatic arterial spasm just as it had done in the earlier work in the rabbit and in a small number of experiments in the goat. There appears to be no species difference in the effectiveness of the drug. From the clinical viewpoint the most significant findings are of course those obtained in primates.

The Rate of Relaxation.—Graphs were drawn for each vessel studied and on them were recorded in detail the measurements of the diameter plotted against time. Similar records were available for the rabbit and the goat. In these it was also possible to study rates of relaxation in vessels which were respectively smaller and larger than those of the monkey. It was found that: (1) A latent period of ½–3 minutes ensued after the papaverine was applied and before the dilatation began. Its duration was not influenced by the size of the vessel or by species differences, nor did it vary with the degree of spasm. It was no longer in cases of complete spasm than in those where the vessel was only

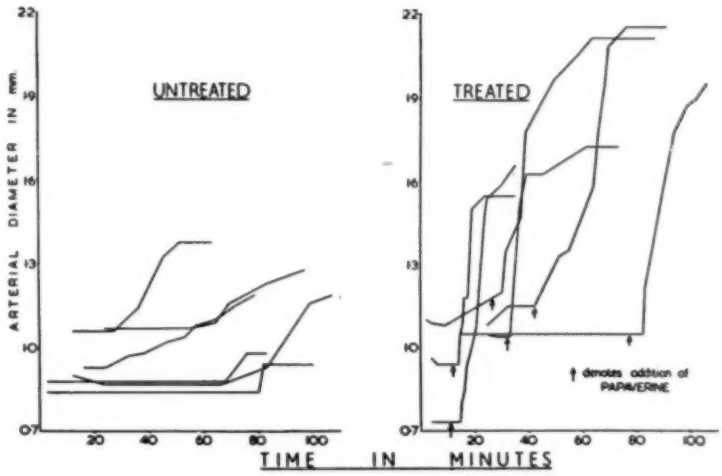

FIG. 210.—Effect of papaverine on spastic arteries in monkeys.

FIG. 211.—Spastic femoral artery of monkey before and after applying papaverine (*see* text).

partly occluded. (2) The rate of relaxation varied with the size of the vessel and the degree of spasm. The fastest to dilate were those in partial spasm, i.e., where there was some visible pulsation. The slowest were the largest vessels in complete spasm. This was found to be true both for the total time taken for relaxation to become complete as well as for the actual rate of relaxation expressed as increase in diameter in unit time.

TRAUMATIC SPASM IN CLINICAL SURGERY

We have been able to collect a number of case reports of patients treated for traumatic arterial spasm by direct application of smooth-muscle relaxing drugs. Some of them were published by different authors, others were kindly reported to us by friends and colleagues. The remainder occurred in our own surgical practice. This collection of cases covers the four years 1952–5. A list of them is given in *Table II*. Papaverine was the drug used in all cases except *Case* 2, in which pethidine was used. In *Case* 9 papaverine was used for a limited period and followed by saline.

Table II.—Clinical Cases of Traumatic Arterial Spasm treated by Topical Application of Papaverine, etc.

Case No.	Reporters	Artery	Result	Type of Case	Drug
1	Sutton and Hadfield, 1952	Brachial	Success	Saline infusion	Papaverine
2	Learmonth, 1953	Superficial femoral	Success	Embolectomy	Pethidine
3	de Takats, 1953	Superficial femoral	Success	Arteriography	Papaverine
4	de Takats, 1955	Superficial femoral	Success	Arteriography	Papaverine
5	de Takats, 1955	Brachial	Success	Fracture	Papaverine
6	Edwards and Lyons, 1954	Brachial	Success	Gunshot wound	Papaverine
7	Spencer and Grewe, 1955	Superficial femoral	Success	Gunshot wound	Papaverine
8	Spencer and Grewe, 1955	Superficial femoral	Success	Gunshot wound	Papaverine
9	Hughes, 1955	Superficial femoral	Failure	Gunshot wound	Papaverine
10	Abbey, 1955	Brachial	Success	Fracture	Papaverine
11	Kinmonth, 1951–4	Brachial	Success	Arteriography	Papaverine
12	Kinmonth, 1951–4	Superficial femoral	Success	Arteriography	Papaverine
13	Kinmonth, 1951–4	Superficial femoral	Success	Arteriography	Papaverine
14	Kinmonth, 1955	Brachial	Success	Arteriography	Papaverine

Comments on the Cases.—

Case 1.—One of us (G. J. H.) was present at the operation and the case was later published by Sutton (1952). The spasm occurred in the brachial artery of a man aged 52 next to the point at which a needle was inserted in the median cubital vein to give a saline infusion. The trauma to the artery appears to have been slight, but the patient was in a condition of wound shock at the time and this possibly increased the irritability of the vessel.

Case 2.—Professor Sir James Learmonth (1953) kindly supplied this report :—

" Man, aged 62, known to have had mitral stenosis for 20 years. At 4.30 p.m. sudden onset of paralysis of both legs while playing golf. Sent to Royal Infirmary, Edinburgh, as saddle embolus of the aorta. Seen at 8.30 p.m. The right leg had recovered all motion and sensation ; the left leg remained paralysed below the knee, with stocking analgesia to the level of the knee-cap. Operation at 10 p.m., under spinal anæsthesia. The embolus occluded the common and deep femoral arteries but did not extend into the superficial femoral. The deep femoral arose 2 in. from Poupart's ligament and the mass of clot extended upwards 1 in. above Poupart's ligament. The three vessels were controlled by tapes and embolectomy was performed in the usual way. When the tapes were finally released the external iliac and deep femoral at once resumed their original size, but the superficial femoral remained in spasm over an area of about ¾ in. While the wound was being sewn up a pledget soaked in pethidine solution, 2 c.c., containing 100 mg. of pethidine (5 per cent solution), was placed around the part of the artery in spasm. In two minutes, when the final stitch was inserted, the superficial femoral had regained its normal calibre except for a small local indentation on the anterolateral aspect, rather like a fishpond fracture of the skull. This seemed to me to show that the pethidine had exercised a local effect and that the drug had not at the time of closure penetrated this part of the wall of the artery."

Pethidine relaxes smooth muscle and we have found that it relaxes traumatic arterial spasm in the rabbit as effectively as papaverine. It has, however, one potential disadvantage : It can be absorbed from the wound and act as a respiratory depressant. On the other hand we could find no evidence of respiratory effects in rabbits when papaverine in doses of 150 mg./kg. or more were instilled into wounds, whereas much smaller doses of pethidine regularly produced apnœa and death under similar conditions. The advised maximum dose of pethidine in man is 200 mg. for an adult. If this limit is not exceeded and if the drug is used on a pledget of gauze, as Sir James Learmonth used it, it appears to be safe and effective.

Cases 3, 4, and 5.—Dr. Geza de Takats (1953, 1956) has kindly sent details of 3 cases of spasm in which papaverine was used. He writes : " During open femoral arteriograms which I often do under spinal anæsthesia just before lumbar sympathectomy twice (out of roughly 120 cases) the artery stopped pulsating after the needle (No. 16 short bevel threaded with polythene catheter) had been withdrawn and firm pressure had been applied. Procaine and hot physiologic saline compresses failed to relieve spasm. Two per cent papaverine solution applied by saturating a small gauze compress started the pulsation." Writing of the brachial artery he says : " In one case of Volkmann's contracture seen in consultation less than 24 hours after injury, exploration and papaverine gave complete restoration of pulse. There was no splinting and no subfascial hæmatoma." De Takats also describes the use of papaverine as an antispasmodic in the iliac arteries during dissection for thrombo-endarterectomy and for relieving the spasm of fresh autogenous vein grafts.

Case 6.—In a case described by Edwards and Lyons (1954) the brachial artery was in spasm following the passage of a bullet through the neighbouring tissues. It was not possible to exclude the presence of thrombus in the vessel until a longitudinal incision was made. It was then sutured and the spasm relaxed by applying papaverine.

Cases 7 and 8.—These were recorded by Spencer and Grewe (1955). In both cases the profunda artery had been shot away and the superficial femoral artery was in spasm, which was relieved by applying papaverine. They also record the occasional complication of spasm in arteries repaired by suture during the Korean campaigns. Papaverine was used to relax it.

Case 9.—Dr. Carl Hughes (1955 a, b) recorded details of a case of arterial spasm in a casualty in Korea where papaverine failed to relieve the spasm and he has also kindly sent us further particulars of the case. The common and superficial femoral arteries were in spasm following a gunshot wound which had destroyed the profunda artery eleven hours before operation. "At surgery the profunda artery was ligated, the spastic segment of the artery dissected free and bathed in papaverine for approximately 20 minutes with little result, following which the artery was bathed in warm saline and the spasm gradually released." It is evident from experimental work and from reviewing clinical cases that spasm is often relaxed more slowly in thicker vessels. It may be necessary sometimes to apply the drug for as long as half an hour before relaxation occurs. Presumably time is needed for the drug to soak into the deeper layers of muscle in the wall of the vessel.

Case 10.—Abbey (1955) records a case in which a 12 cm. length of the brachial artery of a child of 6 years was in a spasm after fracture of the humerus. It regained its full diameter within three minutes of applying papaverine. This rapid response was possibly due to the small artery of a child having a thinner wall for the drug to penetrate and also perhaps because a stronger (5 per cent) solution of papaverine was used than in other cases where the solution was usually one of 2 or 2·5 per cent.

Cases 11–14.—We have encountered 4 cases of troublesome spasm of arteries during open arteriography. Like de Takats' *Cases* 3 and 4, they were infrequent happenings in a large series of arteriographies, but in 2 of them (1 femoral, 1 brachial) the spasm occurred at the beginning and the arteriographic injection could not be completed until the spasm was relieved. An example was *Case* 14, a girl aged 19 years. The brachial artery was exposed in its middle third. It contracted to a white pulseless cord over a length of 2½ in. before any injection could be made. An attempt to introduce the needle below the spastic segment resulted in further contraction. A 4 per cent solution of papaverine sulphate was dropped on to the vessel and then a pledget of gauze containing the solution applied. In six minutes the vessel had dilated sufficiently for the arteriogram to be completed. No further spasm occurred.

Comparison with Results before the Introduction of Topical Drugs.—It is interesting to compare the results of treatment of arterial spasm in this new series with those reported before the use of smooth-muscle paralysing agents was introduced. In 1951 two of us (J. B. K. and G. J. H.) collected and reviewed the published cases of traumatic arterial spasm up to that date (Kinmonth, 1952). The earlier reviews and bibliographies of Cohen helped us greatly (1941, 1944). The brachial or femoral arteries were involved in 37 of the reported cases and the results of treatment of spasm at this site are compared in *Table III* with the later series.

Table III.—Clinical Records of Spasm. The Use of Topical Drugs compared with Other Methods

	Brachial		Femoral		Total		
	Success	Failure	Success	Failure	Success	Failure	Failure
Before 1951	18	8	9	2	27	10	per cent (27)
1952–5 papaverine, etc., used	6	—	7	1	13	1	(7)

The cases recorded as failures are those in which gangrene or ischæmic contracture of muscle ensued. In the earlier series many different forms of treatment were used ranging from simple decompression through irrigation with saline or serum to peri-arterial stripping or excision of the vessel. When the recorded cases were reviewed in 1952 (Kinmonth) the figures showed that the results were worse when stripping or resection was practised. In a few cases procaine solution, usually in low concentration, was applied, probably with the aim of paralysing hypothetical sympathetic vasoconstrictor nerves in the adventitia of the vessel. Procaine has also a direct smooth-muscle relaxing action and if used in effective strength might be expected to have some relaxing effect on the spasm.

The number of successful cases in the first series was swollen by the inclusion of some in which spasm was relaxed by simple decompression. This tends to show the results in a more favourable light than in the later series, in which we have included only the more severe cases where decompression alone failed and further measures were necessary to release the spasm.

It will be seen that there were 37 cases with 10 failures in the first, compared with 14 cases with 1 failure in the second series.

DISCUSSION

The clinical treatment of traumatic arterial spasm is not one which lends itself to a trial by parallel controlled series. The nearest which can be attained

to this has been done in the laboratory experiments and the clinical reviews which have been recorded here. Several questions remain to be solved. One of the most puzzling is the varying irritability of arteries in their response to trauma. Under apparently similar conditions spasm will occur in one subject and not in another and even in one limb but not in its opposite fellow. Another question is the reason for the slower rate of relaxation of larger vessels. Is this due to the drug taking longer to soak into the deeper layers of muscle or is it due to some other factor perhaps to do with the movement of the muscle-fibres over each other as they relax and elongate?

There are perhaps theoretical grounds for using more concentrated solutions of papaverine than the 2·5 per cent which has been the standard one. A stronger solution might have a more rapid effect on obstinate spasm in thick-walled vessels. The hydrochloride cannot be dissolved in concentrations greater than 2·5 per cent, but the sulphate is much more soluble. Solutions of 4 per cent and 5 per cent were used without harmful effects in *Cases* 10 and 14 (*Table II*) and in *Case* 2 a 5 per cent solution of pethidine was used, also without harm. On the other hand we have found in laboratory studies that hypertonic solutions, such as 5 or 10 per cent papaverine sulphate, inserted into wounds in the rabbit caused necrosis to develop in the thin skin of the thigh near the suture line. The vessels, nerves, and deeper structures suffered no harm. It seems best, therefore, to restrict the use of the stronger solutions which are hypertonic to those cases where the spasm is obstinate and slow in relaxing under the effect of the standard solution. It would be advisable also to use the pledget method so that contact of the solution with the skin is avoided.

A complication of the relief of arterial spasm which sometimes requires foresight and attention is the occurrence of intense œdema in the limbs following restoration of the circulation. It is most marked when the ischæmia has been prolonged and in one case (*Case* 9 in *Table II*) caused the loss of the limb. The œdema fluid may collect in fascial compartments to such an extent that decompressing incisions may be necessary.

Practical Considerations.—Most of the clinical recommendations based on the earlier experimental studies still stand (Kinmonth, 1952) and have been confirmed by subsequent work. There are some details which can be amplified and so the following somewhat general principles can be laid down for dealing with a case of artificial spasm.

1. Early exploration is advisable when there are signs of ischæmia following an injury, either closed or open, in the region of a large artery. The most important signs are absence of the peripheral pulses and pallor and coldness of the limb. Muscle is particularly sensitive to ischæmia, and if deprived of its blood-supply for a very few hours becomes irretrievably damaged. Exploration may reveal arterial compression, in which case the artery will resume its normal size and pulsation as soon as blood-clot is evacuated and the tension relieved by incision of fascia and overlying tissues. Nothing further need be done to the vessel. In such a case it is important to secure hæmostasis in the wound and avoid suturing it tightly. At the other extreme of injury the vessel may be found to have sustained organic damage, and excision with perhaps replacement by a graft may be necessary. This will depend on the degree of injury, the importance of the vessel, and the amount of injury in or pressure on collaterals.

2. If the vessel is found to be in spasm it is covered by a 2·5 per cent solution of papaverine hydrochloride or sulphate. It is important to ensure that no particles of blood-clot adhere and that the solution comes into close contact with the vessel. It is also important to expose the entire length of the spastic segment so that none of it, particularly proximally, is left untreated.

3. Should the vessel show no alteration in twenty minutes then a stronger solution may be used. This is best applied on small pledgets of gauze applied to the vessels so that none of the hypertonic solution may damage the skin edges. Our present practice is to keep stocks of 2·5 per cent and also 5 per cent papaverine sulphate solution in the theatre in readiness for emergency use.

4. When the spasm is relieved, hæmostasis is effected, and the wound loosely sutured, it is wise to insert a small drain as oozing is not infrequently caused by the dilating effect of the drug upon the small vessels.

5. Intravenous injections of vasodilator drugs or sympathetic blocks cannot affect the spastic main vessel. Theoretically they may dilate collaterals, but often these too are damaged or compressed and cannot respond. Such measures should not be allowed to delay exploration.

6. The systemic blood-pressure must be maintained at a normal level when necessary by blood transfusion in order to help re-expansion of the vessel and to diminish the peripheral vasoconstriction of wound shock.

7. The rapid restoration of arterial supply to a limb which has been ischæmic for some hours often leads to progressive œdema which may be sufficient to stop the blood-supply once more through the pressure of exuded fluid inside fascial compartments. A similar thing is seen after release of a tourniquet which has been left in place for a prolonged period. Elevation of the limb helps to avoid it, but signs of such an occurrence must be watched for and decompressing incisions of fascial compartments may become necessary. Danger may also result from swelling under encircling bandages or plaster-of-Paris casts.

SUMMARY

1. Traumatic arterial spasm in primates may be relieved by direct application of smooth-muscle relaxing drugs.

2. Papaverine is found to do this as effectively in the monkey as in other species. There are no systemic effects observed and the wounds heal normally.

3. There is a short latent period after applying the drug and before dilatation starts. It is the same for vessels of differing size or in different degrees of spasm.

4. The rate of relaxation by drugs is slower in larger vessels and those in more complete spasm.

5. Reports of 14 clinical cases of arterial spasm treated by topical drugs have been collected and are discussed. The spasm was successfully relieved in 13 of these cases.

Acknowledgements.—We are indebted to many for help in this work. Particularly to Sir James Paterson Ross, Dr. F. A. Simeone, and Sir James Learmonth for their advice. To Mr. Leslie Hall for his help with anæsthetics. To the surgeons who sent us reports of cases. To Mr. M. P. Curwen for statistical advice. To Mrs. H. V. Morgan and Miss D. Aitken for technical help, and to the Photographic Department of St. Bartholomew's Hospital for the photographs and figures.

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SOLITARY CYSTS OF THE RETROPERITONEAL REGION

BY P. W. HOUGHTON, WORCESTER

FROM that group of diverse new formations which may inhabit the retroperitoneal tissues a well-defined type of monocyst may be selected having kindred features with cysts not seldom found within the broad ligaments of the uterus, alongside the vagina, and in proximity to the duct system of the male genital organs. A cyst-candidate for this denomination must necessarily be unilocular, its walls thinly fibrous and its contents clear or mucoid fluid sometimes made iridescent by crystals of cholestrin in mature examples. The cavity may be lined by a single layer of epithelial cells, perhaps ciliated and flattened, or it may be devoid of any cellular lining and this naked interior has been reckoned to be due to centrifugal intracystic pressure. There will be no vascular pedicle or bond of attachment to neighbouring organs save only the tenuous connective tissue conveying nutrition through the scantiest of blood-vessels.

CASE REPORTS

Two patients were seen in September, 1954, each suffering from a retroperitoneal monocyst, the considerable size of one exceeding that of any similar case chronicled in the literature.

Case 1.—Mr. A. V. V., aged 36, a Dutchman, complained that, his belly enlarging equally in rate and size with that of his gravid wife, he became the subject of ocular comment, especially so when the increase continued after her delivery. Paradoxically, there was some loss of weight and a complaint of pain in his left loin. Examination made plain a tense, dome-shaped abdomen and a bulging left flank, both transmitting a fluid impulse, suggesting a diagnosis of a retroperitoneal cyst or a giant left hydronephrosis. *Fig.* 212 portrays the X-ray appearances of the abdomen, the distal colon overlying the cæcum and ascending colon in the right abdominal quadrants. The excretory pyelogram (*Fig.* 213) demonstrates a moderate left caliceal dilatation and the whole picture is remarkably like the illustration in Howarth's case (1950).

OPERATION (Sept. 30, 1954).—The swelling was exposed through a left oblique, subcostal extraperitoneal incision under general anæsthesia administered by Dr. F. W. Parke. The left paravertebral fossa contained an extraperitoneal simple serous cyst of large proportions; its polar regions touched the diaphragm above and the pelvic floor below, and its equator stretched from the left flank to the right iliac fossa, carrying the small bowel and its mesentery far to the right in the cœlomic cavity. The distal colon was ribboned across the cyst and the left ureter was unexpectedly adherent to the cyst instead of the peritoneum. When its boundaries were more or less defined its contents were aspirated and its wall removed, the only intimate attachment lying within the bifurcation of the left common iliac vessels. The ureter now lay unattached along its course from the pelviureteric junction to the pelvic floor, but anxiety felt for its survival was needless since later pyelographic studies provided a picture of normal anatomy (*Fig.* 214). The volume of fluid removed was 1 gall. 1 pt. (5·1 l.).

His convalescence was uneventful.

HISTOLOGY (*Fig.* 215).—The cyst wall is composed of fairly dense collagen with variable amounts of capillaries. There are neither muscle nor glandular elements, nor can any lining be made out.

FIG. 212.—Pre-operative radiograph showing descending and pelvic colon displaced into right flank.

FIG. 214.—Post-operative excretory pyelogram : the left kidney appears normal.

Two larger retroperitoneal deposits of fluid have been recorded by Bayers (1892) and Aldous (1911), but in each case the fluid was an accumulation of bile from a leak through an extrahepatic biliary duct.

As one operates to remove such a vast reservoir the capability of the body for adaptation may cause surprise until the mammalian ability to accommodate the products of conception is recalled.

FIG. 213.—Pre-operative excretory pyelogram : the left kidney appears normal.

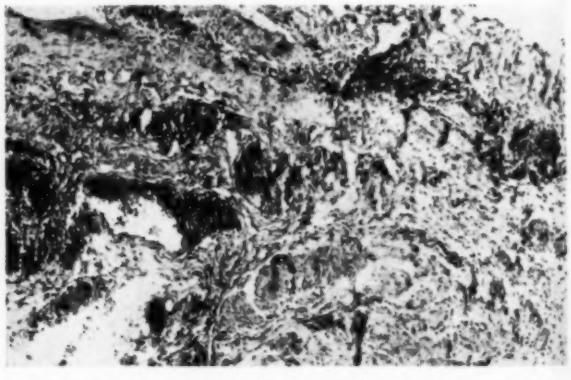

FIG. 215.—Photomicrograph of cyst (Mr. A. V. V.). There is no epithelial or endothelial lining.

Case 2.—Mrs. V. B., a married woman, was referred by Dr. J. Campion Williams, of Worcester. The patient had complained of a dull pain in the right iliac fossa for more than two years and when she presented herself for examination her physician palpated a smooth, mobile, oval cystic tumour in the right flank and he diagnosed a retroperitoneal tumour. Six months previously this patient had been delivered of an anencephalic monster.

On examination, a swelling the size of a grape-fruit lay in the lower right abdominal quadrant. The excretory pyelogram was normal. The swelling was explored through an oblique extraperitoneal incision. External pressure delivered the cyst into the wound, whence it was removed after dividing a few connective tissue adhesions. There was neither anatomical connexion nor relationship with the genito-urinary organs. The wound was closed and recovery was unhindered by complications.

PATHOLOGICAL REPORT.—A tense, thin-walled cyst containing clear, viscid, mucoid fluid. The internal lining is quite smooth.

HISTOLOGY (*Fig.* 216).—The cyst wall is composed of loose, moderately cellular fibrous tissue. The internal surface is covered by a single layer of tall columnar cells.

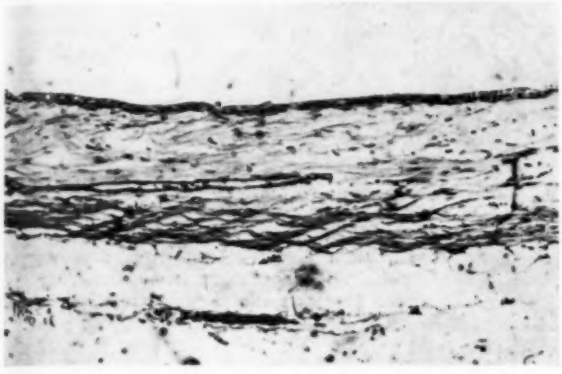

FIG. 216.—Photomicrograph of cyst (Mrs. V. B.). The cyst is lined by a single layer of columnar epithelium.

Two points of difference may be remarked upon. First, tension in this cyst was far greater than in the larger one, yet the lining epithelium was not atrohpic, suggesting that its absence in such a cyst is not a result of internal pressure but is due to a fundamental difference in origin. Secondly, the fluid in the smaller cyst was mucoid—a property derived from the epithelium—but the fluid in the larger, unlined cyst was merely watery, like a transudate.

DISCUSSION

Retroperitoneal monocysts are an abiding nuisance to their host and eventually cause serious trouble by their size. The unlined cysts are innocence itself but they do not cease to increase their dimensions. They should be removed by an extraperitoneal approach without delay.

The origin of these cysts has exercised the minds and, indeed, the imaginations of a succession of surgeons who have chanced upon them, but the primal source remains uncertain. They have appeared in reviews of retroperitoneal cysts and tumours since the classical analyses of Moynihan (1897) and Handfield-Jones (1924) and the names of Harrington and Ganshorn (1939) and Lahey (Lahey and Eckerson, 1934) may be remembered, together with Herdman (1953), who published the largest consecutive series of retroperitoneal tumours in this country and provided a bibliography. Grister (1939) has separated the unilocular retroperitoneal cysts from the omental and mesenteric cysts.

Their origin is sometimes ascribed to mesonephrogenic or Wolffian tissue; the epithelial-lined cysts (*Fig.* 216) may well be wraith-remnants of an ephemeral urinary system and have much in common with certain types of broad ligament cysts and with the cysts found alongside the testes and in the spermatic cord, which derive almost certainly from mesonephric or Wolffian remains (Wakeley, 1943).

Occasionally the epithelium is ciliated and Keith suggests this endowment presupposes a source in the Wolffian duct. It may be observed in passing that polycystic disease of the epididymis is not to be included in the group of paragenital cysts developing from these remains, but is perhaps engendered from faulty union between the efferent ducts of the testes and the rete testes in a manner similar to the cause of polycystic renal disease.

Minor Nichols (1947) has clearly distinguished between the lined and the unlined cysts, an example of each type having been described in the present case reports. The unlined cysts he calls "retroperitoneal cystic lymphangiomata" (*Fig.* 215), because of the absence of epithelium, the serous contents, the occasional presence of cavernous lymph-spaces in the cystic wall, and the equal incidence in the sexes. Although lymphatic dilatations elsewhere are likely to be multilocular, they are sometimes found within the mesentery as large unilocular cysts (Macnab and Menzies, 1950), and this hypothesis is attractive.

This idea of lymphatic origin may be true in another sense, for there is a resemblance between these serous cysts and that "small, closed supradiaphragmatic mesothelial cavity, the infracardiac bursa" (Hamilton, Boyd, and Mossman, 1954), a relic of the right pneumato-enteric mass—a resemblance close enough to imagine another island of embryonic peritoneal membrane as the source of the simplest of all the retroperitoneal cysts.

SUMMARY

Two cases of retroperitoneal monocysts are reported. Their origin and natural history are discussed.

My thanks are due to Dr. F. Kurrein for providing reports on the histology of these cysts and their photomicrographs.

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THE MANAGEMENT OF VOLVULUS OF THE SIGMOID COLON

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VOLVULUS of the sigmoid colon, forming 1–4 per cent of all intestinal obstructions (Hamlin, 1952), is not a sufficiently common condition in this country for any individual surgeon to reach a satisfactory conclusion on its management from his personal experience. At the same time its incidence is large enough for every surgeon to be confronted with a number of cases in the course of his career. The original purpose of this paper was to suggest some tentative conclusions on its management, and to suggest particularly that the judicious use of a rectal tube might substitute elective for emergency surgery. In the course of preparing the paper, however, the writings of Bruusgaard (1947), based on a personal experience which no British surgeon can hope to rival, were found to give authoritative support to the idea of conservative management in the acute stage of sigmoid volvulus, and this paper is now presented in the hope that the method, which is in no way new in theory, may be more widely used in practice.

CLINICAL MATERIAL

A brief account will now be given of 5 personal cases, followed by a summary of the relevant information from a total of 28 cases, 23 having been collected from hospital records. All the cases referred to were treated in the West Middlesex Hospital, the Bristol Royal Infirmary, or Weston-super-Mare General Hospital, between 1946 and 1955.

Case 1.—A male of 69 years was admitted to hospital on Feb. 2, 1951, with a week's history of abdominal colic, constipation, and mucoid discharge from the rectum. During the previous two years he had had two operations for the relief of sigmoid volvulus, so that the diagnosis was not difficult to make, and an immediate Mikulicz resection of an enormous, hypertrophied sigmoid loop was performed. His progress was excellent, and, after closure of his colostomy, he was discharged on April 5.

Case 2.—A man of 82 years, admitted on Dec. 28, 1951, with a similar clinical picture, of twelve days' duration. He had undergone operations for volvulus of the sigmoid colon in March, April, and October, 1951, in Canada. In this case, also, a Mikulicz resection of a distended, hypertrophied sigmoid loop was performed, and, after closure of his colostomy, he was discharged on Jan. 27, 1952.

Case 3.—A man, aged 32 years, who was admitted on Jan. 12, 1955, with 36 hours' history of severe abdominal colic and sacral backache, with absolute constipation. He had had similar attacks at intervals for two years, not requiring surgery. The diagnosis of sigmoid volvulus was confirmed by straight radiography of the abdomen, and at operation he was found to have a generalized redundancy of colon, and an enormous, twisted sigmoid loop. In view of his relative youth, the volvulus was merely untwisted, and he was discharged on Jan. 22 with arrangements made for him to return in April for resection and anastomosis of his sigmoid colon.

On Feb. 22, however, he returned with a further acute volvulus. A rectal tube was passed, with immediate relief of pain and distension, the tube being left in situ for 48 hours. On March 3 a formal resection and anastomosis of his sigmoid colon was performed, and he was discharged well on March 17.

Case 4.—A woman, aged 82 years, admitted on Feb. 17, 1955, with abdominal and sacral pain, constipation, and a mucoid discharge from the rectum, for a week. She had had an operation for relief of sigmoid volvulus in 1952 and showed considerable asymmetrical distension of the abdomen, and a single distended loop of bowel in a straight radiograph. She was fibrillating and had chronic bronchitis, and was most unsuitable for surgery. A rectal tube was passed, with immediate relief of pain and distension, and after a normal bowel action next day she was discharged.

Case 5.—A man, aged 89 years, a patient in a geriatric ward, with congestive failure and left hemiparesis. He was seen after 48 hours of abdominal pain and distension and complete constipation. A diagnosis of sigmoid volvulus was confirmed radiologically, and, as he was manifestly unfit for surgery, a rectal tube was passed, with relief of his symptoms. Six hours later the patient died.

At post-mortem he was found to have a grossly redundant sigmoid colon, still containing some excess of gas and fluid and retaining a half twist. The loop was œdematous and hæmorrhagic, especially at the site of the twist, but was entirely viable.

Summary of Twenty-eight Cases.—

Ages: Over 50 years, 20 cases; under 50 years, 8 cases. Range, 23–89 years.

Average length of history on admission: Over 50 years, 7 days; under 50 years, 2½ days.

Cases with a gangrenous loop: 2 cases (7 per cent). Both under 50 years; both died.

Mikulicz operations: 14 cases, with 3 deaths (22 per cent); 1 gangrenous loop.

Operative untwisting of the loop: 11 cases, with 3 deaths (27 per cent).

Blind cæcostomy: 1 case who died (moribund patient with gangrenous loop).

No operation: 2 cases, with 1 death.

Cases with more than one operation for sigmoid volvulus: 7 cases (25 per cent).

Cases with diagnosis made clinically and/or radiologically: 10 cases, of which 5 cases were known to have had previous operative treatment of a sigmoid volvulus.

DISCUSSION

In the cases reported above, all deaths occurring in hospital are recorded, although many were attributable to associated cardiovascular or respiratory disease, present before operation. From this analysis certain points of importance emerge: Cases with gangrene are few (7 per cent) and carry a high mortality; Pool and Dunavent (1951) give 55 per

cent as the mortality in gangrenous cases. The gangrenous loops were both in patients of the younger age group, in whom the condition is less common but more acute, the average length of history on admission being 2½ days, as compared with 7 days in the older group. The younger patients appeared commonly to have an underlying abnormality in the size and fixation of the colon, and certainly volvulus is a well-recognized complication of megacolon. Several younger patients, including 2 of the fatal cases, were transferred from mental hospitals. The impression that volvulus of the sigmoid colon is a more acute and hazardous disease in younger than in older patients is shared by Dean and Murray (1952), but Hilton and Waugh (1951) suggest the opposite view. Several cases in the present series demonstrated the remarkable time for which a sigmoid volvulus can exist, especially in the elderly, without major vascular changes in the loop.

In any discussion of the treatment of volvulus of the sigmoid, the recurrence rate associated with any procedure less radical than resection of the loop must be considered. The present series includes no systematic follow-up, as the cases were too scattered for it to be attempted, but nevertheless 25 per cent of the cases are known to have had more than one operation for the same condition. Dean and Murray (1952) found that 6 out of 10 patients whose volvulus of sigmoid had merely been untwisted at operation required further emergency surgery. Bloodgood (1909) reported a patient who had 32 recurrences in 16 years. Hilton and Waugh (1951) give the recurrence rate of simple untwisting as 20 per cent.

DIAGNOSIS

Clearly, no satisfactory selection of a method of treatment for a particular case can be made unless an accurate clinical diagnosis is made first. In only 10 of the 28 cases in this series was such a diagnosis made, and in only 5 of 21 cases with no previous surgical treatment for volvulus was a firm and correct conclusion reached pre-operatively. Usually these cases are diagnosed as large-bowel obstructions probably due to neoplasm, and operation is undertaken with the intention of constructing a colostomy.

It is hardly necessary to review the whole clinical picture of volvulus of the sigmoid, but it may be worth while to mention the points which suggest the diagnosis, as a confident diagnosis can commonly be made if the condition is remembered. Pain may not be severe in many, but in some it is more intense and prostrating than is usual in a typical large-bowel obstruction, and this is particularly common in younger patients with shorter histories. The association of abdominal pain with sacral backache in a patient with obstructive symptoms is also suggestive of the condition, the backache presumably being due to pull on the pelvic mesocolon. In patients with short histories and severe pain, the degree of distension is often remarkable for the length of history, and in these the distension is usually asymmetrical and a distinct palpable loop may be felt arising from the pelvis. In less acute cases the degree of distension may be quite extreme, and the presence of a single loop difficult to recognize, but the surgeon is often impressed by the excellence of the patient's general condition in view of the long history and extreme distension (Dean and Murray, 1952). In several cases it has been observed that, after the onset of complete obstruction, the patient continues to pass a little watery mucus from the rectum, and in the absence of a rectal growth this is a suggestive feature. If the diagnosis of sigmoid volvulus is seriously entertained, a straight radiograph

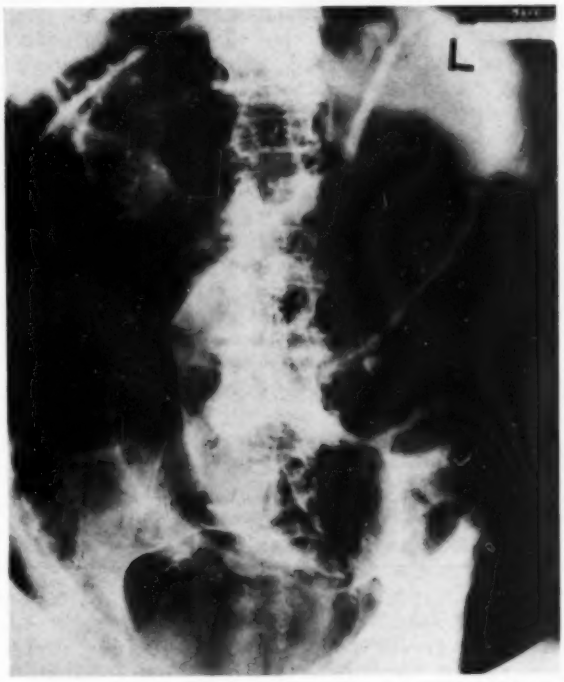

FIG. 217.—The X-ray appearance in a case of volvulus of the sigmoid colon. The film was one belonging to *Case* 5.

of the abdomen is most valuable. The single, enormously distended, loop, arising from the pelvis, is highly characteristic: often, it contains a single fluid level, and it is surprising how long the volvulus may be present before the proximal bowel distends sufficiently to give multiple fluid levels. A typical example is shown in *Fig.* 217, which is the film belonging to *Case* 5.

MANAGEMENT

Two important considerations govern the management of a case of volvulus of the sigmoid colon: the frequency with which the condition is seen in patients of advanced age and poor physique; and the high incidence of recurrent volvulus, where any procedure less radical than resection of the sigmoid loop is employed. The available methods of treatment are:—

1. Operative untwisting of the volvulus.
2. Resection of the sigmoid loop by the Mikulicz operation.
3. Resection and end-to-end anastomosis of the sigmoid loop.
4. Reduction of the volvulus by rectal tube, with or without subsequent resection.

1. Operative Untwisting of the Volvulus.—This is the simplest operative method available, and, as an emergency surgical procedure, has the merit of being the minimum necessary to save life, of

providing absolute certainty of diagnosis, and of giving the surgeon secure knowledge of the viability or otherwise of the twisted loop. On the other hand, it is not free from hazard, and carried a 27 per cent mortality in the present series, a high figure due to its employment in the worst risk cases. When simple untwisting is carried out, the chance of recurrence is high, and Pool and Dunavent (1951) advocated resection and anastomosis six to twelve weeks later, but when an elderly person is convalescent from an emergency operation, surgeon and patient are equally reluctant to embark on a further major operation to resect the redundant sigmoid loop. Hence the matter is not pursued further until the patient returns with a recurrent volvulus, when the problem arises again in its original form. Nevertheless, simple untwisting of the volvulus remains the method of choice in bad-risk cases, when relief by a rectal tube fails, or clinical doubt as to the viability of the loop can only be resolved by laparotomy.

2. Mikulicz Resection of the Sigmoid Loop.—This is probably the most satisfactory operative method in acute sigmoid volvulus, since it both relieves the condition and eliminates the risk of recurrence. It is sometimes said that the mortality is prohibitive (Aird, 1949), but the present series does not really support that view. Leaving out 1 case, with advanced gangrene of the sigmoid loop, the mortality of immediate Mikulicz resection of the twisted segment was 15 per cent, which is not unreasonable in the type of patient usually encountered. Certainly, if the condition is discovered unexpectedly at laparotomy for large-bowel obstruction, and in cases where non-operative reduction has failed, Mikulicz resection should be carried out, if the patient is a reasonable operative risk. In cases with a non-viable loop it is the only practicable procedure, although the chances of success are poor, while in elderly patients it may be an attractive alternative to resection and anastomosis, when an elective operation is performed following non-operative relief of acute volvulus.

3. Resection, with End-to-end Anastomosis of the Sigmoid Loop.—This method would not commend itself to many as a suitable technique in acute sigmoid volvulus, but Dean and Murray (1952) employed it in 11 such cases, with only 1 death. It is clearly the best method, however, for a young patient following non-operative relief of an acute volvulus, as described in *Case* 3, while it is also applicable to such of the elderly patients as present good operative risks.

4. Reduction of the Volvulus by Rectal Tube.—In the present series there are 4 examples of this method, used in different ways. In *Case* 3 the patient was subjected to one unnecessary operation for untwisting a sigmoid volvulus, but on his second admission the successful use of a tube made it possible to perform an elective resection and anastomosis of his sigmoid loop, with an entirely satisfactory result. In *Case* 4 decompression with a rectal tube was employed in a case in which the risk of any operative procedure was unduly high. The patient continues to be subject to the risk of a recurrence of her volvulus, but the risk is no greater than it would have been with operative untwisting of the loop, and a major operative hazard has been avoided. In *Case* 5 the patient would certainly have died on the table had operation been attempted, but by use of a rectal tube the need for operation, and a sequel in the coroner's court, was averted. In this instance reduction was not quite complete, and the tube should undoubtedly have been left in position and not immediately withdrawn. In only 1 case, among the 23 collected from hospital records, was a rectal tube employed, and in that instance under a misapprehension. A patient suffering from a recent cerebrovascular accident was admitted with a large-bowel obstruction, and was diagnosed as a carcinoma of the colon. A rectal tube was passed, with complete relief of the distension, but during the next four days signs and symptoms recurred. A rectal tube was again passed, with complete relief, but was immediately followed by operation to construct a colostomy. A completely reduced sigmoid volvulus was found, and the abdomen closed, but the patient died a few hours later. Operation might have been avoided in this case had it been realized that it is very improbable that a rectal tube could be passed through a malignant stricture tight enough to hold up flatus.

Bruusgaard (1947) suggests that, once volvulus has occurred, the main bar to spontaneous reduction is the rapid development of great distension of the loop, which cannot untwist because of its size, but is likely to do so if emptied. In 136 cases in which intubation was attempted, 123 were fully relieved, and recovered; 3 died despite complete relief of the volvulus, and fully viable loops, as shown at post-mortem; in 1 case the loop was perforated by the tube, and this patient died, despite immediate operation; and in 9 cases a tube could not be passed, and operation had to be undertaken.

Hamlin (1952) treated 11 cases by attempted intubation, and failed once. Isaacson and Greer (1952) treated 7 cases by intubation, and advocated leaving the tube in situ for two or three days, and undertaking definitive surgery in two or three weeks. All three authorities passed their tubes under vision, using a sigmoidoscope or a long proctoscope, but in the 3 cases described in this paper a stout rectal tube was passed without difficulty, with the patient in the left lateral position and a finger in the rectum guiding the tube upwards. There is no doubt when the manœuvre is successful, as there is a large and prolonged escape of flatus, followed by liquid fæces, and accompanied by a dramatic relief of abdominal distension.

In all cases treated by intubation reasonable certainty must exist of the viability of the loop, and, while vascular changes appear to be infrequent and late, any major degree of abdominal tenderness should suggest the possibility, and laparotomy be considered. If the tube is successfully passed, it can easily be secured by strapping or a stitch and retained for 48 to 72 hours. After this a decision can be taken whether radical surgery is indicated in the individual case, and whether it should take the form of a resection and anastomosis or a Mikulicz operation. The patient can be suitably prepared for a bowel resection, and an unhurried operation under optimum conditions can be substituted for a less satisfactory emergency procedure.

SUMMARY

An account is given of 5 personal cases of volvulus of the sigmoid colon, and a total of 28 cases is analysed to show, in particular, the high incidence of recurrence, with any but radical surgery, and the mortality of various operative procedures.

Some points in the diagnosis are discussed.

The management of acute sigmoid volvulus is reviewed, and the valuable place of conservative treatment of the acute case, with subsequent elective surgery, is stressed.

My thanks are due to Mr. W. J. Ferguson and Mr. J. Scholefield, of the West Middlesex Hospital; to Mr. W. R. Black, of Weston General Hospital; and to Professor R. Milnes Walker and the surgical staff of the Bristol Royal Infirmary, for allowing me to use material relating to cases under their care.

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INCOMPLETE AORTIC VASCULAR RINGS

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MALFORMATION of the aortic arch and its branches producing tracheo-œsophageal compression has emerged in recent years as a definite syndrome in the infant, but its presence in the adult as a cause of such compression phenomena has not attracted as much attention. It is becoming increasingly clear, however, from the growing literature on this subject, that a fair percentage of such anomalies produces symptoms in adult life, often of a chronic and even bizarre nature.

A complete double aortic arch may produce no symptoms (Mathey, Facquet, Alhomme, and Combaz, 1952), but when it does it gives rise to serious tracheo-œsophageal compression of a comparatively acute type, declaring itself in the first few days or weeks of postnatal life. This is a fairly rare anomaly. Gross (1953) quotes 21 cases operated upon by him since his first case in 1945.

An incomplete aortic vascular ring is, however, more common and may remain symptomless for years. Sometimes it is discovered during routine X-ray examination. Mathey and others (1952) believe that it remains latent in about two-thirds of the cases. In the other third, it usually declares itself in the latter half of adult life, possibly owing to changes in the size of the aorta and its calibre.

The normal aortovascular configuration is achieved by a development process which converts the fourth left branchial arterial arch into the adult left ascending arch of the aorta. The right fourth branchial arch forms the innominate artery and the proximal part of the right subclavian artery. The fifth aortic arches are rudimentary, inconstant, transitory vessels which atrophy at an early date. The pulmonary arteries are formed from the sixth pair of aortic arches, the right sixth arch loses its original connexion with the dorsal aorta. However, on the left a portion of the sixth arch remains as a large vessel connecting the dorsal aorta with the pulmonary artery. This is known as the ductus arteriosus [botalli]. Complete vascular rings, as found in newborn infants, are mostly due to the persistence of the right branchial arch, producing a bifid aortic arch, the anterior limb of which passes in front of the trachea and the posterior, larger one, usually behind the œsophagus. Tracheo-œsophageal compression follows in the majority of such infants, with growth of mediastinal structures and elongation of the body.

An incomplete vascular ring is mostly associated with a right aortic arch. A right aortic arch is sometimes found with other cardiac anomalies, but often occurs alone. The comparatively unknown extracardiac anomaly of an incomplete double aortic arch is not clinically manifest in many cases, though the existence of such incomplete rings may be demonstrated by suitable roentgenological and other studies. The two components of the incomplete vascular ring are the right aortic arch on one side and various degrees of persistence of the 4th and 6th branchial arches representing a rudimentary, mostly fibrous, left aortic arch and ductus arteriosus. Through the attachment of the ductus (or ligamentum) arteriosus to the left pulmonary artery, a peri-tracheo-œsophageal ring is produced, often constrictive in nature. Additional constrictive elements may be contributed by the great vessels coursing through the tracheo-œsophageal mediastinum (mostly in front, sometimes behind, rarely between). Mathey and others (1952) describe three types of such incomplete double aortic arches.

A right aortic arch *per se* will not produce any symptoms unless restrained by an opposing incomplete band on the other side. Such pressure may not only affect the tracheobronchial mediastinum, but also the right main bronchus and often the recurrent laryngeal nerves. In all such cases the recurrent nerves are epibronchial, passing under the right arch and left ligamentum arteriosum, and may be affected by pressure on either side. The pressure on the œsophagus is mostly on the right side and behind, while tracheal compression is mostly anterior and on the left. A diverticulum of the aorta may produce

posterior œsophageal pressure even without an incomplete ring.

Incomplete aortic vascular rings can also occur in infants and produce acute symptoms and death. In point of fact, one of the earliest suggestions for division of the ligamentum arteriosum as therapy

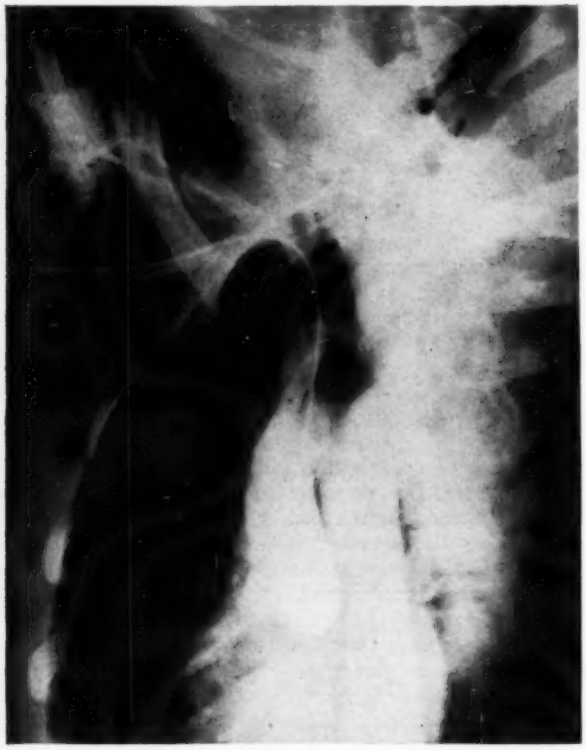

FIG. 218.—Retrograde aortogram showing the right aortic arch and its four branches.

in such cases was made by Gross (1953), after studying the autopsy findings in a 4-months-old baby. Very rarely a mirror image of this usual type of incomplete double arch is found, with partial repression of the right arch. Here a normally placed left aortic arch is associated with a right-sided ligamentum arteriosum causing pressure. The diagnosis is naturally difficult.

Symptomatology is normally vague and diverse. Dysphagia is the commonest symptom, but never marked. Regurgitation and postprandial distress is often present. Respiratory symptoms are less important—though dyspnœa, specially of a paroxysmal nature, may occur. Cough is normally unproductive, unless bronchial compression is present, and may be hoarse in character as a result of recurrent nerve compression. Pain may be present, but mostly follows ingestion of food and is high dorsal or substernal in location.

Diagnosis is practically dependent on investigations, especially radiography. Plain chest radiographs show tracheal compression, which is better shown by laminograms. But the essence of diagnosis lies in a barium study of the œsophagus, which is pushed forward and to the left and anteriorly by a smooth, rounded, often pulsatile shadow, prevertebral in location—the common situation being opposite the fourth dorsal vertebra. The aortic arch is seen on the right (except in the very rare mirror image type—situs inversus), but the cardiac configuration is usually within normal limits.

Pulse and pressure differences in the arms are often present, but are not diagnostically significant. The finding of a right aortic arch, pulsatile posterior indentation, and forward displacement of the œsophagus at the upper dorsal level is almost diagnostic, though other possibilities such as intrathoracic goitre, new growths, mediastinal nodes and cysts, and saccular aneurysm of the aorta have to be considered and ruled out. Here angiography is of the greatest value. Intravenous angiocardiography may not produce sufficient radio-opacity in the aorta and its branches for accurate study, but retrograde injection of dye through a polythene catheter introduced from a radial (or brachial) artery gives excellent delineation (*Fig.* 218). This should be carried out in both P.A. and R.A.O. positions. In a case of inverse (mirror-image) incomplete double aortic arch, the L.A.O. position is important. In such rare cases, the presence of the left-sided aortic arch at first excites no suspicion, but the posterior displacement of the œsophagus should suggest the diagnosis and the need for a retrograde angiographic study.

CASE REPORT

HISTORY.—M. L. B., aged 30, male, a railway official, was admitted to the K.E.M. Hospital, Bombay, on Sept. 22, 1953, complaining of dysphagia and attacks of breathlessness of 20 days' duration. The dysphagia was progressive and at the time of admission he could only swallow liquids. Occasional regurgitation of food was present. The attacks of dyspnœa (choking) lasted for a minute or two and came on intermittently, mostly at rest. Occasional attacks of retrosternal (behind and below the jugular notch) discomfort and pain were also present.

Previous medical history revealed nothing of interest except an attack of dysentery a year ago. No history of syphilis. Father died of coronary thrombosis at the age of 50.

CLINICAL EXAMINATION.—A well-developed and nourished man of about 30 years with good colour and no clubbing of nails. Pulse 86 and regular. Respiration 18 per min., normal. A few small lymph-nodes were palpable in the neck.

Respiratory System: Trachea in the midline. A tracheal tug of moderate force was felt. No other abnormal findings.

Circulatory System: No clinical abnormality was detected in examination of the heart. Peripheral activities were normal. Blood-pressure right arm 110/70, left arm 100/70.

Other systems were essentially normal.

Examination of urine, stool, blood (hæmogram) showed nothing abnormal. The Kahn test was negative.

Indirect laryngoscopy showed the right cord fixed in cadaveric position. The left cord showed compensatory over-activity.

Œsophagoscopy revealed an extra-œsophageal pulsatile tumour at the level of the aortic arch, situated posteriorly and to the right of the œsophagus.

Radiography (fluoroscopy): Lung fields clear, diaphragm moving well, trachea in midline, right-sided aortic arch. A barium swallow showed a posterior and right-sided indentation of the œsophagus by a mass at the level of the fourth dorsal vertebra. The extra-œsophageal mass was pulsatile. Heart was essentially normal.

Radiographs confirmed the fluoroscopic findings (*Figs.* 219, 220).

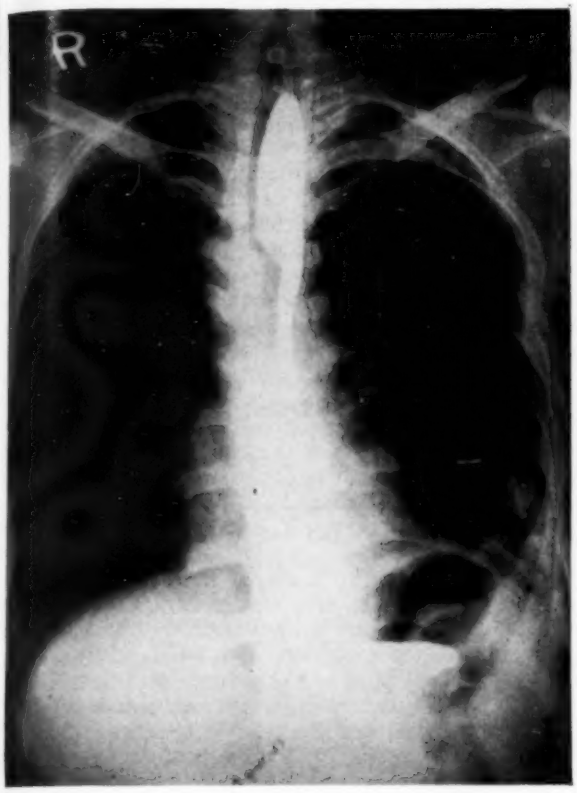

FIG. 219.—P.A. radiograph with barium swallow, showing well-defined 'cut-out' produced by the aneurysmal dilatation of incomplete arch. Such posterior 'cut-outs' if seen pulsating under the fluorescent screen are diagnostic of a vascular ring anomaly.

FIG. 220.—Right anterior oblique view of the patient with barium swallow. Œsophagus is pushed forwards with considerable compression, causing dysphagia.

Tomograms showed some anteroposterior compression of the trachea.

Retrograde aortography done by introducing a polythene catheter into the aortic arch through the left brachial artery, at the elbow, and injecting 15 c.c. of dye ("Iduron" Cilag, 70 per cent iodine) revealed a right-sided aortic arch, right subclavian, right common carotid, left carotid, and left subclavian arteries. The left carotid was seen crossing in front of the trachea, but the left subclavian arose from the posterior part of the right aortic arch, from a diverticulum, which appeared to form the main extra-œsophageal mass compressing the gullet from behind. (*See Fig.* 218.)

DIAGNOSIS.—A diagnosis of an incomplete double aortic arch with a right aortic arch and a right descending aorta and a left ligamentum (or ductus) arteriosum was made. The compression element was obviously the arterial ligament and therefore it was decided to explore the thorax from the left side.

OPERATION (P. K. S.).—On Nov. 30, 1953, under endotracheal gas–oxygen (with pentothal and curare) anæsthesia (Dr. G. S. Ambardekkar) the chest was entered through the bed of the fourth rib on the left side. A right-sided aortic arch was seen with disposition of great vessels as normally seen with a usual left-sided aortic arch (except that the right subclavian and carotid arteries arose separately, close together, and there was no innominate artery). The left common carotid was coursing obliquely in front of the trachea from its origin from the right arch to enter the neck on the left side, and the left subclavian arose from a pouch-like diverticulum of the right aortic arch passing behind the œsophagus and compressing it. A left ligamentum (or ductus) arteriosum was present, around which the left recurrent nerve was hooked and stretched (*Fig.* 221). This band,

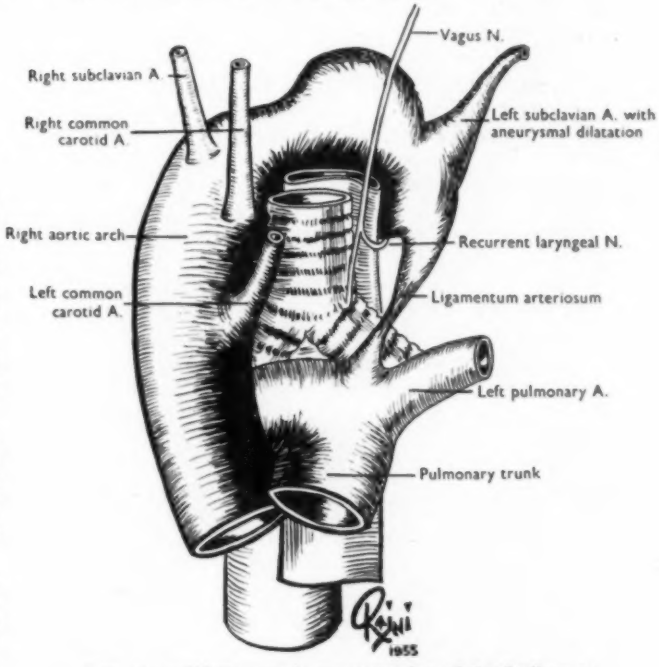

FIG. 221.—Diagrammatic representation of the findings at operation. Note the saccular aneurysm at the root of the left subclavian artery.

which appeared partly patent, was the main constricting agent and was divided between Pott's toothed clamps and the small lumen and closed with a continuous suture of 5/0 arterial silk. The chest was closed in layers after expanding the lung with an underwater-seal drain in the left eighth space.

PROGRESS.—In the immediate post-operative period severe hoarseness amounting to near aphonia developed,

apparently due to the handling of the left recurrent nerve (the right recurrent being already involved), but this cleared up in a few days. The voice improved rapidly and was almost normal in pitch four weeks later, when an indirect laryngoscopy showed both vocal cords to be moving freely and well. Dysphagia cleared up almost immediately following the operation and except for occasional and slight difficulty in swallowing liquids in small quantity, he was free from symptoms when he was discharged from the hospital on Dec. 30, 1953.

Fourteen months later, follow-up studies indicate that the patient has no symptoms, and on examination fully functioning vocal cords, no hold-up of barium in roentgen studies of the œsophagus, and no evidence of tracheal compression. The rounded, pulsatile cut-out on the posterior wall of the upper thoracic œsophagus is still seen, as also the displacement produced by the right aortic arch, but evidently there is little interference with the passage of a bolus from this. There have been no attacks of retrosternal discomforts or pain since the operation.

DISCUSSION

The part played by vascular anomalies in the production of obstructive changes relating to œsophageal or tracheal obstruction is not widely appreciated in the field of thoracic diagnosis except in pediatric surgery, where the subject has been high-lighted by acute obstructions in the newborn or in early postnatal life.

In adult life the appearance of œsophago-tracheal obstructions most frequently suggests superior mediastinal compression of a neoplastic nature, or in later life aneurysmal dilatations of the aorta. The possibility of vascular anomalies producing such symptoms is small in adult life. Mathey and others (1952) in an analysis of such cases mention only two instances treated surgically. Gross and Neuhauser (1951) reported 40 cases of vascular anomalies causing compression of the trachea or œsophagus in children since 1945 (Gross, 1945), while during the same period Gross has had only 4 such cases in adults. It therefore appears that there are some types of vascular anomalies, at least, which either escape detection in infancy or do not produce symptoms until later (about the third or fourth decade). Changes taking place in the aorta around this age are probably responsible for the beginning of symptoms, as also the comparative fixity of the chest wall and emphysematous changes in the lungs. While in the child or in the newborn infant symptoms are usually produced by complete or incomplete duplication of the aortic arch, in the adult it appears that the symptoms of constriction follow from an incomplete double arch anomaly with a ligamentum arteriosum or a ductus arteriosus as one of its components.

The diagnosis is in no way different from the condition in the infant. Any posterior compression of the œsophagus in the upper mediastinum is suspicious, and if the possibility is borne in mind it may be diagnosed, perhaps, with greater frequency than it is now. In the case reported here it is significant that the symptoms were of very short duration. This indicates that a major anomaly may exist for many years before producing constrictive symptoms which may even take an acute form. The recovery of the laryngeal palsy following the left ligament division brings out the probable truth in the observation made by Mathey and others (1952) that in those vascular anomalies of the aortic arch both the recurrent laryngeal nerves hook around either the aorta and/or the duct and therefore are intrathoracic in origin. In addition to the bands which complete the constricting ring, it must not be forgotten that other anomalies often coexist, such as diverticula, often amounting to aneurysmal dilatation of the aortic arch or its branches, and sometimes to abnormal transit of the blood-vessels themselves (dysphagia lusoria) (Gross, 1946).

When such additional anomalies are present they must be treated individually and on their merits, for example, the posterior compression may be relieved by slinging up the aortic arch as advised by Gross and Neuhauser (1948) or by division of the aberrant vessels (Gross, 1946). Such division can only be done if the vessel is an aberrant subclavian artery.

The results of division of incomplete vascular arches in adults are not as dramatic as in similar conditions in infants. However, in those cases where such anomalies are beginning to produce symptoms in adult life, division of the constricting band is not only rational but imperative. The mere presence (as an accidental finding on roentgen examination) of a vascular anomaly is not enough to warrant surgical exploration unless obstruction and compressive phenomena are present and symptoms produce sufficient discomfort or disability.

The right aortic arch is not too common an anomaly. Probably a fair percentage of people with a right aortic arch have other vascular anomalies as well, but only a small percentage of them develop symptoms. Surgery for incomplete vascular arch in the adult is therefore only indicated as a compulsive and not as an elective measure.

SUMMARY

The literature on incomplete aortic vascular rings is reviewed. The differences between complete rings, which mostly declare themselves as emergencies in the very young, and the incomplete rings, which usually produce symptoms late in adult life, are brought out. Symptomatology and diagnosis of such anomalies are discussed and a report of an incomplete aortic vascular ring with a right-sided arch treated surgically is presented.

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GANGRENE DUE TO ACUTE MASSIVE VENOUS OCCLUSION OF A LIMB

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GANGRENE due to occlusion of the veins of a limb with a completely patent arterial tree is rare. To date, 2 cases have appeared in the British literature (Clain and Nussbaum, 1950; Martin, 1953), while in America prior to 1949 DeBakey and Ochsner (1949) found only 3 reports.

Since attention was first directed to this type of gangrene by Bergeret, Guillaume, and Delarue (1932) about 30 cases have been reported, mainly by continental clinicians. In many of these cases the underlying pathology has been largely a matter of surmise, complete autopsy protocols being few. For this reason the following case is presented.

CASE REPORT

A man, aged 56 years, developed pain and swelling in the left leg while under treatment for bronchopneumonia, a white blood-count at this time being 16,500/cu. mm. No differential count was done. The only relevant past history was of recurrent thrombophlebitis in both lower limbs for the past sixteen years.

At the onset the condition appeared to be a deep vein thrombosis of the usual type. The limb was elevated and heparin therapy started. Later phenylindanedione (Dindevan) was substituted. Three days later the leg and foot were cold and blue. All the peripheral pulses were present. On the fourth day the swelling was more marked and extended to the groin, while all pulses below this level were absent. The skin below the knee was purple, and above it was red and hot. At the junction there were numerous petechiæ. By the ninth day the swelling involved the groin and scrotum, the femoral pulse could not be felt, and there was early gangrene of the toes (*Fig.* 222).

Anticoagulant treatment was continued until the thirteenth day and then stopped with a view to amputation. Before this could be carried out, death occurred on the fifteenth day from pulmonary embolism.

POST-MORTEM FINDINGS.—The arterial systems of both limbs were completely dissected. Apart from mild atheroma and arteriosclerosis the whole system was patent.

FIG. 222.—The swollen left leg with subcutaneous blebs of serosaneous fluid and distal gangrene.

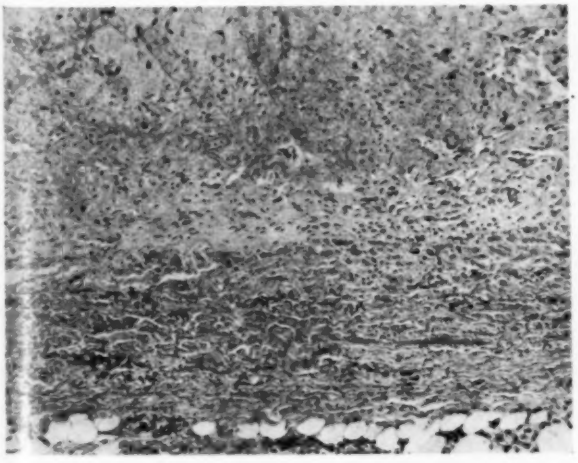

FIG. 223.—Thrombosis of left femoral vein. Early organization. Absence of inflammatory cellular exudation. H. and E. (× 60.)

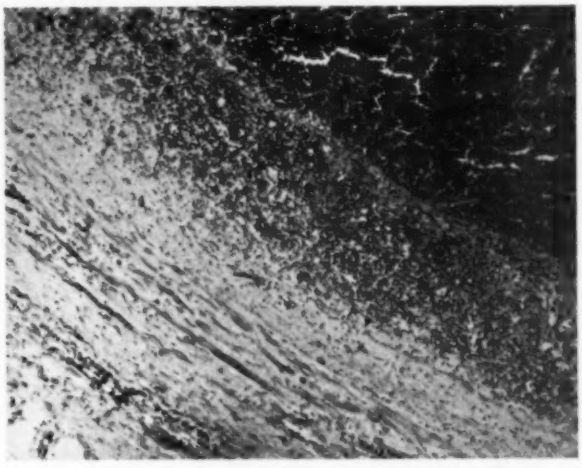

FIG. 224.—Right femoral vein showing phlebothrombosis, and a well-established zone of organization where thrombus remains adherent. H. and E. (× 60.)

The veins of the left leg were completely occluded by well-established ante mortem mixed thrombus. This extended along the iliac veins to the inferior vena cava. The veins of the right leg also contained firm ante mortem clot almost to the inferior vena cava, where the clot ceased abruptly. It had retracted to one side of the wall. The lumen of these vessels therefore was not completely occluded, excepting the lumen of the right internal saphenous vein, where retraction had not occurred.

FIG. 225.—Right internal saphenous vein showing recent complete occlusion. Distortion of lumen and thickening of wall due to organization and recanalization of a previous thrombotic process. H. and E. (× 10.)

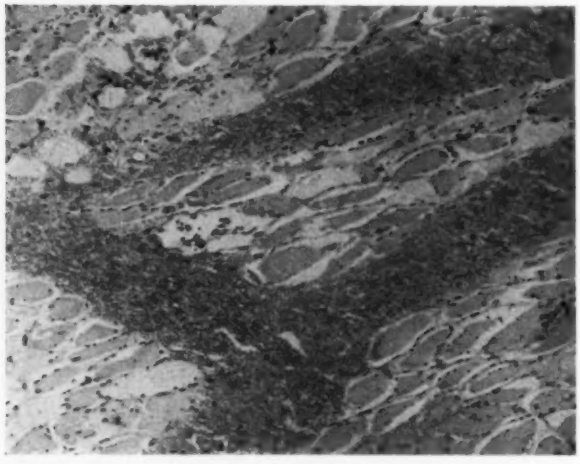

FIG. 227.—Infarction of skeletal muscle of left leg. Interstitial hæmorrhage, œdema, and loss of striation. H. and E. (× 60.)

The muscles of the left calf were firm, œdematous, and hæmorrhagic, suggesting partial infarction. Those on the right side appeared normal.

The other findings included resolving bronchopneumonia with embolic thrombus, almost 12 in. in length, lodged in both pulmonary arteries and extensive propagated thrombus distal to this; an aneurysm of the apex of the left ventricle, which did not contain thrombus; and enlargement of the liver (2,500 g.) and spleen (650 g.), without associated lymph-node enlargement or apparent gross marrow abnormality.

On microscopical examination the limb arteries showed only early atheroma and arteriosclerosis. The veins of the left leg showed the appearances of phlebothrombosis with organization just beginning (*Fig.* 223). In contrast, the thrombus in the veins of the right leg was in a more advanced state of organization (*Fig.* 224). There was evidence that the saphenous vein of this limb had been occluded in part at least on previous occasions, because it showed recanalization and organization of long standing, an observation in keeping with the patient's history (*Fig.* 225).

FIG. 226.—Skin from dorsum of left foot. Early gangrene with intense venous and capillary engorgement. H. and E. (× 60.)

The skin on the dorsum of the left foot showed early gangrene with intense capillary and venous engorgement in the dermis (*Fig.* 226). This is not seen in gangrene resulting from arterial disease alone. Sections from the left gastrocnemius muscle revealed extensive diffuse interstitial hæmorrhage with œdema separating the sarcolemmal sheaths from their contained sarcostyles. The nuclei and the striations of the muscle were lost and the whole showed diffuse eosinophilia due to infarction of venous origin (*Fig.* 227).

Histology of the liver, spleen, mediastinal and abdominal lymph-nodes, and femoral bone-marrow showed the appearances of lymphatic leukæmia. The infiltration of the marrow by leukæmic cells was early but nevertheless distinct, and the infiltration of the other organs was less marked.

DISCUSSION

In the present case, and those described by Grégoire (1938) and Bergeret, Guillaume, and Delarue (1932), the onset was not very dramatic. Indeed, in the early phase these appear to have been cases of phlegmasia alba dolens which had progressed to phlegmasia cærulea dolens as the venous blockage extended, and finally ended in gangrene when the obstruction became complete. Deep violet cyanosis did not quite reach the knee in our case.

Generally, the onset of symptoms is sudden and the pain very severe. Soon after the onset of pain the limb becomes swollen, and a deep violet cyanosis

of the skin appears, spreading in a centripetal fashion from the toes. The upper limit of this distinctive colour may reach the thigh or groin. Petechiæ develop, often most marked at the junction between normal and discoloured skin. Later, blebs of dark serosanguineous fluid form.

Decrease in skin sensation and fall in the skin temperature occur *pari passu* with the colour changes. The arterial pulses are present at first, later becoming imperceptible, the distal ones disappearing first. The symptoms and signs are typical if followed from their inception. If the case is seen first at a late stage considerable difficulty may be experienced in differentiating it from incipient gangrene due to arterial block.

Although loss of peripheral arterial pulsation occurs in these cases this may be due to the concomitant swelling of the part making palpation of the pulses impossible. The complete dissection of the arterial tree carried out in our case suggests that the absence of these pulses by clinical examination does not necessarily mean that the arteries are blocked. It is clear that in some cases differentiation from arterial occlusion is difficult if not impossible, and cases have been recorded where surgical exploration of the limb arteries has been carried out.

Besides the local manifestations there is profound general upset with circulatory collapse. In about 25 per cent of reported cases this has been so severe that death has occurred at this stage. In the remainder, gangrene supervenes and death occurs in about 50 per cent, while, of the survivors, the great majority need major amputation.

Aetiology and Pathogenesis.—The disease is commonest in the third to fifth decades. The upper limbs are rarely involved. While exceptionally both lower limbs may be affected, the condition is usually unilateral, the left leg being affected three times more commonly than the right.

The only element common to most cases is the presence of infection, usually abdominal or puerperal, but occasionally respiratory. Opinion is divided as to the relative importance of venous obstruction and the loss of arterial pulses. Many have attributed this latter to reflex vasospasm from the presence of accompanying thrombophlebitis. This receives some support from the works of DeBakey, Burch, and Ochsner (1939) and Boyd (1947). It is noteworthy, however, that in those cases where specific measures have been taken to overcome this by sympathetic block, or by vasodilator drugs such as papaverine, little benefit has accrued. As already stated, the loss of arterial pulsation on clinical examination may be due to œdema and not to a blocked artery or arterial spasm. From our autopsy findings we believe that the essential lesion is complete blockage of the venous return. This is total in the present case as far as can be ascertained. When venous block is total, blood will be pumped in until the head of pressure in the vascular bed of the limb is equal to that in the arteries and flow will then cease. Provided that this happens quickly and no collateral drainage opens up, then the distal parts of the limb will die. There is experimental proof that gangrene of a limb will occur if there be complete occlusion of the venous return (Fontaine and Pereira, 1947).

In tracing events in the present case, attention is drawn to the following points—the venous systems of both legs and of the pelvis contained thrombus almost to the origin of the inferior vena cava; the coiled embolus in the lungs must have lain originally in this vessel and in the terminal portion of the right common iliac vein; the phlebothrombotic process is of longer duration (estimated at 4–6 weeks) in the right leg compared with the left from the amount of organization and clot retraction. It is visualized that a recent thrombotic process started in the right leg and spread *gradually* up the deep veins. The limb did not swell since time was allowed for collateral veins to open before the iliac veins were occluded. The thrombosis spread into the inferior vena cava, and the left common iliac vein was then occluded *suddenly* at its junction with the cava. A rapid retrograde thrombosis took place in the veins of the left lower limb, perhaps aided by an altered state of coagulability of the blood due to lymphatic leukæmia. No time was afforded for collateral channels to open. In view of the sixteen-year history of ' thrombophlebitis ' it is possible that the venous drainage of the legs was also considerably impaired before this last illness. The final event was detachment of the clot in the inferior vena cava and terminal part of the right common iliac vein producing fatal pulmonary embolism.

Treatment.—If seen early, and the patient's general condition warrants it, an attempt at thrombectomy might be made. However, in view of the present case it is suggested that attention be paid not only to the affected limb but also to its neighbour, since it may be the site of a silent venous thrombosis. Whether or not operation be tried the patient should be heparinized to prevent further obliteration of the venous circulation and as a possible prophylactic against pulmonary embolism. The latter is a not uncommon event. Treatment of possible arterial spasm has not proven of much value, but since spasm of vein wall occurs when there is phlebothrombosis a good case can be made for some form of sympathetic interruption. It would appear more important to heparinize the patient, and to abolish sympathetic influence by heating the rest of the body, by vasodilator drugs and by morphine-induced sleep, rather than by operative interference or by injection of the sympathetic ganglia. Further, since it is obvious that all means to aid the venous return will be helpful, the affected limb should be elevated above an angle of 45° with the body and, if the patient's condition allows it, active exercise of the limb may be tried. In the event of gangrene occurring the usual surgical steps will be called for. It is well to delay amputation for as long as possible since the extent of tissue loss will be much less than appears at first.

SUMMARY

An example of acute massive venous occlusion of the lower limb with resulting gangrene is recorded. The autopsy results are given.

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SUCCESSFUL PARTIAL RESECTION OF THE SMALL BOWEL IN THE CASE OF HÆMOPHILIA

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WITH the ready availability of blood transfusion and greater knowledge of the clotting mechanism, successful major surgery in hæmophiliac patients is being recorded with increasing frequency.

CASE REPORT

HISTORY.—A male, aged 26 years, was admitted to the General Hospital, Birmingham, with a history of colicky abdominal pain of 48 hours' duration. The pain was central at the onset, but had localized in the right iliac fossa after 24 hours and had persisted. Nausea and loss of appetite were present throughout; the patient had vomited once and the bowels had been opened normally.

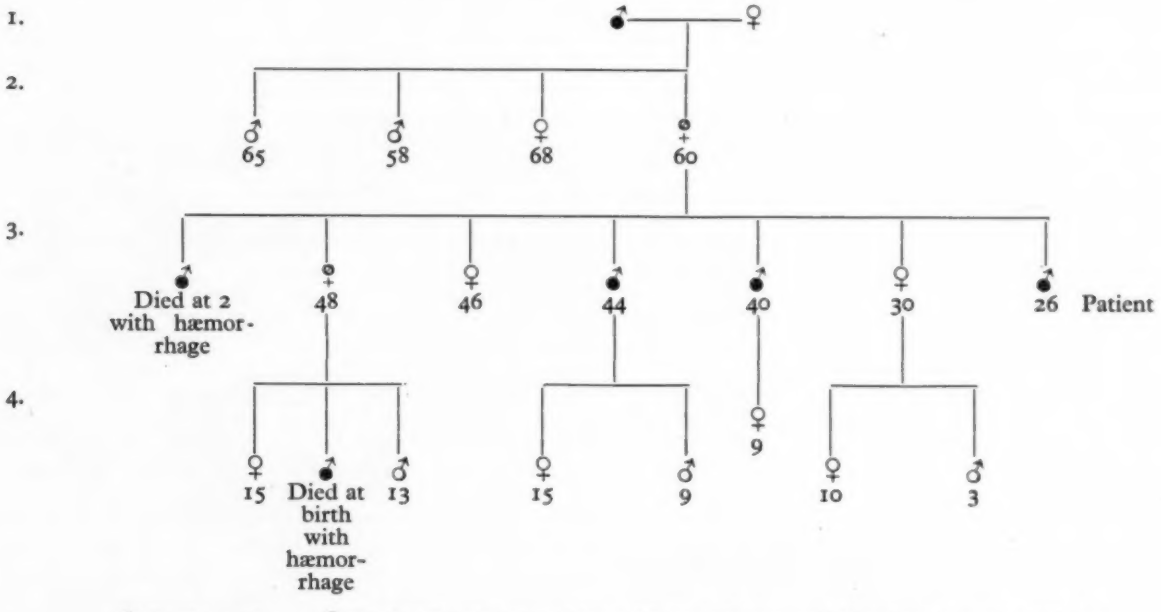
He had been transferred from a local hospital with a diagnosis of 'acute appendicitis in a hæmophilic'. Blood investigations done at that hospital were as follows:

Bleeding time: 3 min.
Clotting time:—
Capillary method 12 min.
Wide tube method 16 min.
Hæmoglobin 80 per cent
Platelets 474,000 per c.mm.
Clot retraction } Normal
Prothrombin time }

ON ADMISSION.—Temperature 101° F. There was now generalized abdominal rigidity which was of the type associated with septic peritonitis rather than with blood in the peritoneal cavity. Bowel sounds were still present though diminished.

Rectal examination revealed tenderness in the recto-vesical pouch. A fairly confident diagnosis of acute appendicitis with generalizing peritonitis was made. In view of the pyrexia and marked abdominal rigidity, hæmoperitoneum was considered unlikely.

PAST HISTORY (obtained subsequently).—The patient had had spontaneous bruising in his muscles all his life—usually the calf muscles. Melæna and epistaxis occurred occasionally and bleeding into a knee-joint once—there being no residual deformity or thickening. He had never vomited or spat up blood, and stated that he did not bleed unduly when he cut himself.



♂ Bleeder male. ♀ Carrier female proved by evidence of abnormal bleeding in male offspring.

FIG. 228.—Patient's family tree.

FAMILY HISTORY (*Fig.* 228).—Both of patient's brothers were similarly affected, i.e., they bruised easily. One brother had had a hæmarthrosis of the knee and one had been in hospital for six weeks following a dental extraction. No girls in the family were affected, but the maternal grandfather was known to be a bleeder. The outstanding feature in all three brothers was the tendency to bleed into their muscles and bowel—they were all familiar with " passing black stools and going weak ".

OPERATION.—An intravenous blood-drip was set up. The abdomen was opened through a right paramedian incision; there was considerable oozing from the wound which could not be completely controlled by diathermy or ligation.

The peritoneal cavity was full of recent blood; a 6-in. length of lower ileum was purple and thickened with bruising, which extended into the mesentery.

The appearance was suggestive of either a mesenteric thrombosis or embolism or an interstitial hæmorrhage into the bowel wall. Arteries were pulsating normally right up to the edge of the discoloured bowel and the gross thickening of its wall suggested the latter diagnosis. No definite bleeding point was found; the appendix was normal. Two feet of ileum were resected, including the affected segment and an end-to-end anastomosis was performed. The blood was carefully removed from the peritoneal cavity. A rapid search for bleeding points revealed none. The abdomen was closed in layers with suprapubic drainage of the pelvis. Section of the specimen confirmed that it was an interstitial hæmorrhage.

PROGRESS.—During the days after operation fresh blood was given as follows: 1st day, 3 pints; 2nd day, 1 pint; 3rd day, 3 pints; 4th day, 1 pint; 5th day, 1 pint; 6th day, 3 pints. No further blood was given. Normal saline, dextrose or Hartmanns' solution were given between the transfusions to maintain the intravenous drip. On the second and third post-operative days the patient developed a mild paralytic ileus which responded rapidly to gastric suction through a Ryle's tube and intravenous infusion. At this stage signs of extreme fear appeared, although morphine gr. $\frac{1}{4}$ was being given 6-hourly; chlorpromazine 50 mg. t.i.d. was therefore given, with excellent soothing results.

On the fourth and fifth days a severe melæna occurred which required further transfusions. On the tenth day he had a mild epistaxis.

Considerable oozing from the wound necessitated frequent changes of dressing and a large hæmatoma formed in the subcutaneous tissues at the wound site, later becoming septic. The skin edges separated, but the anterior rectus sheath remained intact.

The patient was finally discharged on the forty-eighth day.

PATHOLOGICAL REPORT (Dr. D. B. Brewer) (*Fig.* 229).—" Portion of small intestine 55 cm. long showing moderate œdema. 16 cm. from one end was a length of bowel 13 cm. long showing marked interstitial hæmorrhage.

" Microscopically the hæmorrhage appears recent. It is mainly in the submucosa. The mucosa and adjacent muscle is generally normal, but there are areas of necrosis of muscle and smaller areas of necrosis of the mucosa. This mucosal ulceration is related to gross œdema, which is present as well as the hæmorrhage. A mesenteric lymph-node shows reactive hyperplasia with dilated sinuses containing numerous red cells."

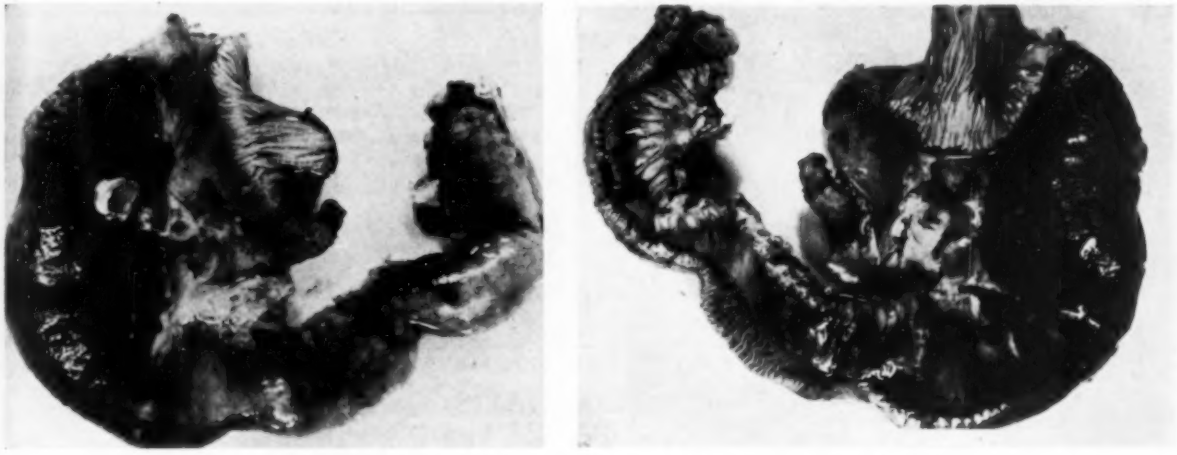


FIG. 229.—The excised portion of hæmorrhagic, œdematous bowel.

BLOOD INVESTIGATIONS.—A preliminary attempt to establish the nature of the bleeding tendency using prothrombin consumption and thromboplastin generation tests was carried out on the eighth day following admission and after 12 pints of blood were transfused. This was done, as not infrequently any deficiency in thromboplastin generation may persist despite transfusion. Nevertheless, the results were equivocal and it was only by re-examining these findings in the light of subsequent tests that some abnormality was seen. Since a diagnosis was desirable the patient's brother attended the laboratory for investigation, the results of which are as follows:—

Hess's test: Negative. Bleeding time: 2 min. Clotting time (37° C.) 10 min. Prothrombin level 100 per cent. Prothrombin consumption 52 per cent.

Thromboplastin generation test: Defective generation using brother's plasma and normal serum. The picture showed a definite deficiency of anti-hæmophiliac globulin, i.e., the brother had hæmophilia.

Three weeks after admission and approximately a fortnight after the last transfusion a further study of the patient's clotting mechanism showed:—

Clotting time (37° C.) 9 min. Prothrombin level 100 per cent. Prothrombin consumption test 40 per cent.

Thromboplastin generation test: The distinctive pattern of anti-hæmophiliac globulin deficiency was revealed. It was thus established that our patient had hæmophilia.

DISCUSSION

Craddock, Fenninger, and Simmons (1948), in a critical review of the subject, state that minor surgery in hæmophilia is commonly carried out with success. In major surgery, however, they stress the distinction between that carried out on exposed sites where pressure or hæmostatic substances can be applied,

e.g., amputation of an arm (Blalock, 1932) or skin-grafting (Davidson and Levenson, 1945), radical mastectomy (Vance, 1939), and internal surgery where local measures cannot be employed. They also lay down four criteria for establishing the diagnosis of true hæmophilia :—

1. Laboratory test showing prolongation of the clotting time. (This should be modified in the light of recent advances, since it is well known that hæmophilia may exist in the presence of a normal clotting time.)
2. A family history of the condition, although not present in every case.
3. A past history typical of hæmophilia.
4. Occurrence in a male.

It is now established that a thromboplastin generation test should be done on all suspected cases.

Several of the case reports in the literature do not fulfil these criteria and may not have been true hæmophilias. If these strict criteria be applied, the mortality-rate for internal operations appears to be between 50 and 60 per cent. In the last few years, however, more successful cases have been reported, viz. :— Appendicectomy (Landry and Shonkimas, 1950), splenectomy for traumatic rupture (Lund, 1950), nephrectomy (Vermooten, 1950), and gastrectomy for bleeding duodenal ulcer (Walker, 1955).

SUMMARY

A case of hæmophilia with interstitial hæmorrhage into the wall of the ileum and hæmoperitoneum is reported. Resection of the affected segment was successfully performed.

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SOME OBSERVATIONS ON LEONTIASIS OSSEA AND OSTEITIS DEFORMANS, PAGET'S DISEASE

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THIRTY early and advanced cases of leontiasis ossea have been investigated and observed for many years. Leontiasis ossea is a focal fibrous dysplasia of the maxilla, which extends to the bones of the face, and after many years involves one or more of the long bones of the skeleton. The nomenclature is confused, and leontiasis was the name given first to the tubercular form of leprosy of the face, and, later, Virchow in 1864 applied it to the enlargement of the bones of the face. The names leontiasis ossea and cherubism are purely clinical descriptions without any pathological basis. Cherubism is of more recent origin. It is the label given to dysplasia of the jaws occurring in infants and children (*Fig.* 230). The morbid anatomy and the normal blood-chemistry are identical with early cases of leontiasis ossea. The cherub-like face is sometimes familial and occurs in more than one member of a family. I have seen 3 cases of cherubism and there are about 6 or more cases recorded. Some of these infants were carelessly fed by the bottle.

FIG. 230.—A photograph of a well-marked case of so-called cherubism. A girl, aged 15 years, abnormality first noticed at 7 years.

Leontiasis ossea has been called hyperostosis (Horsley, 1895), focal osteitis fibrosa, osteitis fibrosa circumscripta, chronic hyperplasia and recently fibrous dysplasia or abnormal development of bone (Ingram, 1951). However, the names leontiasis ossea and Paget's disease have been in use for 90 years and they will continue to be applied in spite of what is written and spoken. Leontiasis ossea presents a smooth, round, hard, painless swelling, or enlargement, of the alveolus of the molar region of the maxilla (*Fig.* 231). The swelling extends to the canine fossa, the nasal process, the roof of the orbit, and to the malar bone. The palatal surface of the alveolus is not involved at first. The antrum is filled by soft spongy bone, leaving a small cavity—the size of a little finger nail—situated immediately below the floor of the orbit. There are specimens in the Museums of the Royal Colleges of Surgeons of England and Edinburgh, and in Charing Cross Hospital, showing this encroachment on the antrum. It is usual for these patients to see a dental surgeon first, often when they are under the age of 10 years and the swelling has been present but

unnoticed for about three years. The majority of the cases of osteodystrophy are segregated to the diagnostic radiography departments and it is said that about 50 cases of Paget's disease per annum are seen in the radiography department of a large London hospital (Fairbank, 1951). The radiograph shows a dense opacity of the body of the maxilla, particularly the floor of the orbit, and in some cases the bone appears to be cystic or porous. In most of the cases the swelling progresses and becomes more dense, the palate increases in width and length, the maxillary tuberosities and the alveolus increase in size, especially in edentulous patients, who, for this reason, require new dentures every three or four years. The enlargement may remain stationary for years, but there are periods at long intervals when it progresses rapidly, and particularly in young patients. As a relatively small area of the skeleton is involved, the blood-calcium and phosphorus and the urine are normal. The nasal cavity is involved at a late stage. The inferior turbinated bones are thickened and prominent. The bone of the normal concave floor of the nose and the inferior meatus are raised into rounded convexities. The antra and frontal and sphenoidal sinuses are almost obliterated by spongy new bone. Three cases developed at a late stage a progressive deafness indistinguishable from otosclerosis. The microscopy of the petrous bone of Paget's disease shows porous new bone of the capsule of the labyrinth and oval window with osteoid tissue and giant cells. The bone changes are similar to those of otosclerosis, but they are more diffuse, more porous, and involve most of the petrous bone. There is more osteoid tissue and giant cells are more profuse. The trabeculæ of bone are broken up and replaced by irregular islets of soft bone. There is absorption accompanied by reformation of soft vascular bone, but in some areas the bone is dense and lamellar. The haversian canals are enlarged and irregular in shape. The marrow is replaced by fibrous tissue. The hæmatoxylin stain of the new bone is a deeper blue, but as the microscope slide becomes older this staining fades. This also is seen in the new bone of otosclerosis and in that of osteogenesis imperfecta with blue sclerotics. The bony changes in otosclerosis are localized to the oval window and the footplate of the stapes at first, but later they may spread to the bony capsule of the labyrinth. A number of cases of deafness indistinguishable from otosclerosis have been associated with blue sclerotics and fragilitas ossium, i.e., osteogenesis imperfecta tarda. Cleminson (1927) in his paper records cases which he illustrates with a microscope section of the petrous bone of a patient with blue sclerotics given to him by Professor Nager, of Zürich. The section shows multiple foci of sclerosis and new bone, one big focus of the promontory and anterior part of the bony cochlea including the oval window and footplate of the stapes and three separate foci around the semicircular canal, the round window, and internal auditory meatus. The bony changes are similar to those of otosclerosis, but the osteoid tissue is almost absent and ossification is more marked. The bone was obtained from a post-mortem of a man 25 years of age who was completely deaf in the right ear and had only a remnant of hearing in the left ear. The pictures portrayed in the microscopical sections of the three diseases can be identified and recognized without difficulty. Knaggs (1926) divided leontiasis ossea into two groups; (1) Creeping periostitis of the bones of the face and skull; and (2) A diffuse, or local, osteitis in which there is complete absence of any periosteal bone. The whole bone appears to be enlarged and Knaggs suggests that the disease commenced in a nasal sinus or in the centre of the bone. I have not seen such a case. All the above 30 cases could belong to the first group of creeping periostitis, and after many years 3 of them developed marked thickening with bosses and tumour-like excrescences of the margins of the orbit and frontal bone. These 3 advanced cases and some specimens with large excrescences, like tumours in the Royal College of Surgeons' Museum, might have been placed in the second group by Knaggs. The pathology of both groups is the same and this division is of no clinical value.

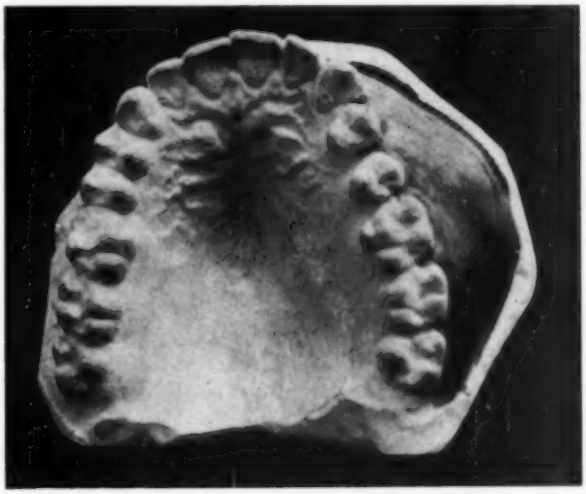

FIG. 231.—Plaster cast of the maxilla of a case of leontiasis ossea taken at the age of 14 years. Swelling first noticed at 7 years. Paget's disease diagnosed at 34 years.

MORBID ANATOMY

Operations for the correction of the deformity are not performed until the age of 21 years, and not before full growth has been attained. The bony swelling cuts like an unripe pear. It is vascular and small cysts about the size of a large pin's head may be seen. Larger cysts have been found on rare occasions. The periosteum is thickened and vascular and this vascularity and engorgement with blood is the cause of the warmth and local rise of temperature of the long bones of Paget's disease. It is not an inflammatory process (*Fig.* 232). The typical fibrous dysplasia is seen under the microscope. Giant cells or osteoclasts are numerous in early cases and in young patients. These cells tend to disappear as ossification progresses. Osteoblasts under the periosteum and areas of ossification and calcification are seen. The formation of new bone and its absorption by osteoclasts is continuous and proceeds simultaneously.

A compensatory ossification to give rigidity to the normal soft bone is usual. The formation of new bone in layers to form a mosaic or lamellar

arrangement is seen in Paget's disease. A study of the specimens of Paget's disease in the Museum of the Royal College of Surgeons shows the medullary cavity of the long bones to be reduced in size by spongy bone. Six out of the 30 cases observed by the writer for twenty years or more proved to be Paget's disease, particularly of the calvarium. The remaining 24 cases were observed only for variable

FIG. 232.—Microscopy of a section of the maxilla of a boy of 12 years. It shows the fibrous dysplasia seen in leontiasis ossea and Paget's disease. The bone of older patients shows larger areas of new-bone formation and its absorption.

short periods of about five years. The predominant pathology of leontiasis ossea and Paget's disease is the replacement of bone by fibrous tissue, a dysplasia or excess of osteoid tissue which leads to the softening and loss of rigidity, hence the compensatory enlargement and the deformity of the bones. There is a rapid decalcification and the bones affected react by the formation of a fibrous dysplasia followed by ossification. At the same time the existing bone is absorbed by osteoclasts and new bone is laid down under the periosteum by the osteoblasts. The new-bone formation is greater than the destruction of bone and this process is in excess of the normal deposit of new bone and its absorption. The bone destruction and bone formation proceed simultaneously with a patchy distribution. The cause of this dysplasia is obscure and unknown, but it is assumed that the metabolism of the ossification and development of the bone is defective. The deposition and absorption of calcium and phosphorus are disturbed. The nutrition and even the vitality of the bones are inhibited, hence the general description or name of osteodystrophy. The reason for the increase of alkaline phosphatase in the blood-serum and in the affected bone of Paget's disease is not understood. but it is known that phosphatase is associated with calcification and the formation of bone. Phosphatase is an enzyme capable of liberating inorganic phosphorus from the organic glycerophosphates and esters of the tissues. It is most active in a *p*H 9 which does not exist *intra vita* because a *p*H below or above 7·4 is incompatible with the normal function of the body. The estimation of the *p*H of bone-marrow is difficult and unreliable. It is said that the presence of magnesium accelerates the action of phosphatase, but excess of magnesium retards calcification. Phosphatase is present in growing bone and depends on the activity of the osteoblasts. It is found in the developing and growing mammary glands, also the intestines, the prostate, pancreas, kidneys, etc., tissues which do not ossify except under rare pathological conditions. The increase of phosphatase and the diminution of the phosphorus of the blood-serum is stated to be diagnostic of hyperparathyroidism, but the analysis of the bone of Paget's disease reveals the same relation of phosphatase and phosphorus.

The diagnosis of leontiasis ossea is not difficult, and it is usually obvious, but in its early stages the question of a tumour arises. A tumour is circumscribed, the alveolus is expanded, and both its plates present almost a symmetrical swelling. Leontiasis ossea usually affects the outer plate of the alveolus, and it has been mistaken for a fibroma or osteoclastoma. A fibroma or osteoma is confined to one bone and does not spread to the other bones of the face. Sometimes the pathologist gives a diagnosis of fibrosarcoma on the microscope slide only.

Paget's disease, or osteitis deformans, is a more generalized, more diffuse, fibrous dysplasia, with areas of decalcification of the bone. Fibrous dysplasia is the result of the reaction of bone to rapid decalcification. The bones affected and the distribution of the dysplasia varies. It sometimes commences in the skull and face then extends to the long bones, but more often it affects the spine and pelvis, the long bones, and extends to the calvarium of the skull. A marked increase in the cementum of the teeth is said to be diagnostic of Paget's disease, but many patients are edentulous when seen for the first time. It is stated that leontiasis ossea differs from Paget's disease because it is discovered at an earlier age and that the long bones are not involved in leontiasis ossea. It is true that leontiasis ossea is diagnosed earlier because it is more easily seen and it is more evident. It is said that Paget's disease of the spine and pelvis is not recognized before the age of 40, but radiography has now shown that it commences at an earlier age. The long bones of cases of leontiasis are involved at a later period than in Paget's disease, but they are sooner or later affected, and Knaggs records 4 such cases in his book. In young subjects Paget's disease has been monostotic like leontiasis ossea. Both diseases are identical, but they appear to differ in the bones affected. The morbid anatomy and the negative biochemistry are similar, the blood-calcium and phosphorus are normal, but the phosphatase in Paget's disease and in leontiasis ossea is increased. Fragments of the normal bone of the calvarium and of the clavicle were obtained from the autopsies of 12 adult patients. The fragments were weighed, and the amounts of calcium and phosphorus were estimated by the Courtauld Institute of Biochemistry, Middlesex Hospital. The amounts of calcium and phosphorus varied and they did not conform to any sort of pattern or uniformity. It appeared that the calvarium contained more calcium and phosphorus than the clavicle, and the shaft of the

clavicle more than that of its head or epiphysis. Old people sometimes had more calcium and phosphorus than the younger adults. Variations in the amounts of calcium should have been expected, because it is known that the skeleton is the main store of calcium needed for the body. In wasting diseases and starvation the calcium of the bone is much diminished and atrophy follows. The average amounts of calcium and phosphorus found in the bone ash of the 12 post-mortem specimens were :—

Calvarium : Calcium 18 per cent, Phosphorus 11 per cent, 18 g. per 100 g.
Head of clavicle : Calcium 11 per cent, Phosphorus 5·3 per cent.
Shaft of clavicle : Calcium 17 per cent, Phosphorus 10 per cent.

The amount of calcium was always greater than that of the phosphorus, but the ratio of calcium to phosphorus in normal mammalian bone is 1·99 to 2·04 and it is constant. These estimates were compared with that found in specimens of bone obtained from an operation biopsy of Paget's disease of the maxilla in a woman of 40, and secondly that of a post-mortem specimen of the femur of a man with well marked Paget's disease.

1. Maxilla—Woman, aged 40	2. Femur—Male, aged 62
Calcium 7 per cent	14 per cent
Phosphorus 0·1 per cent	2·5 per cent
Alk. phosphatase 750 K.A. units	590 K.A. units
Magnesium 0·6 per cent	1·2 per cent

As controls the maxilla of a woman of the same age and normal bone and the normal calvarium of the man with Paget's disease were analysed and the following results were obtained :—

1. Calcium 21 per cent	2. 19·1 per cent
Phosphorus 22 per cent	2·3 per cent
Alk. phosphatase 5 units	10 units
Magnesium (not estimated)	1·0 per cent

It will be seen that the phosphatase of the diseased bone is definitely increased and the phosphorus is diminished in proportion. The amount of phosphatase, calcium, and phosphorus found in bone is greater than that of the blood-serum. The magnesium content was unaffected.

It is acknowledged that the number of specimens obtained from Paget's disease is insufficient and more are required to arrive at any definite conclusion. Unfortunately, the opportunities for the examination of fresh post-mortem specimens and biopsies taken during an operation are very rare.

AETIOLOGY

The direct cause of Paget's disease is obscure, but more is known of the contributory or indirect causes. Hereditary disposition is difficult to establish because very few patients know anything of the diseases of their grandparents. There is evidence that a disposition is inherited (Galbraith, 1954). Fragilitas ossium with blue sclerotics is hereditary and familial. Traumatism is doubtful. Some patients give a history of a blow or injury in cases of leontiasis ossea. It occurs in both sexes. There are 23 females and 7 males in the above series. Sepsis is not a cause and no evidence of an inflammatory origin has been established. The bone is sterile. Septic teeth and sinusitis do not cause leontiasis ossea but the affected bone is very easily infected and necrosis may occur after an operation, or after the extraction of teeth. The Wassermann reaction and evidence of syphilis are undoubtedly negative. There is no evidence that Paget's disease is associated with an endocrine disorder. It is known that the administration of pituitary growth hormone produces an increase of the weight and dimensions of the skeleton, but it does not affect or hasten the maturation of bone. Acromegaly patients with enlargement of the pituitary gland develop a subperiosteal overgrowth of the bones of the mandible, face, hands, and feet. The parathyroids are believed to regulate the blood-calcium. Excessive doses of parathyroid hormone raise the level of the calcium in the blood and lower the level of inorganic phosphorus and of alkaline phosphatase. When the calcium of the blood exceeds the normal 10 mg. per cent the excess of calcium is excreted in the urine and fæces. Similarly, the excess of phosphorus above 4 mg. per cent is excreted as calcium and other phosphates. It is suggested that vitamin D_2 acts by stimulating the parathyroids. The hyperplasia of one parathyroid body and not all four suggests that there is some other factor, perhaps the interaction of pituitary and parathyroid hormones. Moreover, in some cases of fibrocystic disease of bone the parathyroid bodies are normal. The morbid anatomy, blood-chemistry, and clinical course of hyperparathyroidism with fibrocystic disease of bone differs very considerably from Paget's disease. The destruction of bone is greater than its reconstruction in fibrocystic disease.

On the other hand Paget's disease is the reverse, the reconstruction and new-bone formation is greater than the destruction. Giant-celled tumours and an increase in the giant cells occur in fibrocystic disease. Patients with concretions and calcification in the kidneys or giant-celled tumours require an investigation for hyperparathyroidism even though fibrocystic disease of the bones may not be discovered. It is probable that the bone is affected later than the nephrolithiasis. The diminution of the inorganic phosphorus and an increase of the alkaline phosphatase of the blood is a characteristic of hyperparathyroidism (Baird, Grainger, and Rowlands, 1954). Three of my cases of Paget's disease were given injections of parathyroid hormone with disappointing results. One of these patients, a woman of 45, was admitted to the hospital suffering from a rapid progression of the disease and the blood-chemistry was negative. She was given daily injections of parathyroid hormone for three weeks, but after this time the injections were discontinued because her symptoms increased and the disease progressed more rapidly. The injections did not relieve her pain. Hormones of the sex organs have an influence on the ossification and development of bone. Continued administration of testosterone for long periods produces a hyperossification. The male hormone stimulates ossification and the female hormone counteracts and retards ossification (Cooke, 1955).

Nutrition.—Nutritional defects are well recognized causes of the osteodystrophies such as rickets and osteomalacia. Calcium deficiency and nutritional defects occur during pregnancy, starvation, and severe renal disease. It is accepted that the skeletons of birds and mammals act as a reservoir for calcium and phosphorus, which can be drawn upon when these minerals are deficient or inadequate in the diet.

During the pre-laying period hens store calcium and phosphorus in the medullary bone of the skeleton and the calcium to phosphorus ratio is increased to a minimum of 4·25. When the hens are laying and on a low calcium diet the loss of calcium and phosphorus from the skeleton is increased and the loss of phosphorus is slightly less than that of the calcium. Rapid changes occur in the marrow-bone during the egg-laying cycle and intense bone formation alternates with bone destruction. When the intake of calcium is normal and when active shell formation is not taking place the lost calcium is restored. The greatest proportion of the losses of calcium was in the ribs, sternum, ileum, ischium, pubis, and coccygeal vertebra (Taylor and Moore, 1954). The bone changes seen in the laying hen give some idea of the mechanism of the calcification of bone, and feeding experiments on hens may give more information. Vitamin D_2 (calciferol) is said to increase the absorption of calcium and phosphorus. Excessive doses of vitamin D_2 raise the calcium and phosphate levels of the blood and then the phosphates in the urine are increased. Mellanby (1954) has shown by experiments on puppies fed on phytin, i.e., calcium magnesium phytate, that the administration of vitamin D_2 increased the absorption of the calcium up to a certain level. Phytic acid inositol hexaphosphate, present in some cereals such as oatmeal, combines with calcium to form phytin which is not utilizable by man. Phytic acid hinders the absorption of calcium by combining with it and rendering it unavailable. However, phytic acid does not account for the decalcification of bone in Paget's disease. Excess of phosphorus prevents the normal absorption of calcium and insoluble calcium phosphate is excreted by the intestine and also by the acid urine, hence the fall in the blood-calcium. The ratio of calcium to phosphorus is about 1 : 2 under normal conditions. Animals remain healthy with a ratio of 1 : 5, but 1 : 14 produces bone abnormalities. This ratio is important. Deficient phosphorus or a ratio of calcium of 14·8 : 1 phosphorus produced marked rickets. The influence of calcium, phosphorus, and vitamin D_2 is being investigated. It is accepted that leontiasis ossea and Paget's disease arise from a disorder of the calcification and ossification of bone. There is evidence that osteodystrophy is produced by a deficient diet. Glock and others (1937) showed that osteofibrosis was produced in goats fed for a relatively short period on a ration consisting of flaked maize and bran with a minimum amount of hay. Swellings of the jaws and lameness occurred and the blood-serum calcium was low. Brittle bones and osteoporosis occur in dairy cows when there is a mineral deficiency in the feeding. Rats given drinking water containing ten times the average amount of sodium fluoride developed white brittle sclerosed bones and a disturbance of kidney function. Excess of calcium and phosphorus in the diet of animals produced sclerosis of bone. Goats and pigs on a diet deficient in calcium and phosphorus, or if there is an excess of phosphorus, develop dysplasia of the jaws. The late Sir Arnold Theiler (1934) started a programme of experimental feeding of common farm animals to elucidate the problem of osteodystrophic disease. Feeding experiments on goats were carried out by Groenewald and others (1940) at Onderstepoort, South Africa. A low-calcium and high-phosphorus diet for fifteen months led to a diagnosis of osteofibrosis, but later atrophy and osteoporosis were found. Loss of appetite and intermittent diarrhœa aggravated the condition. Groenewald came to the following conclusions :—

1. Starvation proves to be an important factor in the development of bone atrophy in kids which received a diet low in calcium and high in phosphorus. The ratio of calcium to phosphorus is wide.

2. Clinical symptoms of rickets are observed when the phosphorus intake is low and that of calcium high.

3. When the calcium and phosphorus intakes are both low, with a normal ratio, histological examination of the bone shows rickets.

4. The inhibition of normal growth due to semi-starvation results in the development of bone atrophy and not osteofibrosis. Even rickets, which is more easily produced than osteofibrosis, in time gives way to atrophy.

Theiler states that rickets and osteomalacia result from phosphorus deficiency and that calcium deficiency produces osteofibrosis. The phosphorus content of the feeds is as important as that of calcium. The condition of the bones during feeding experiments is ascertained by biopsy and radiography. Dogs given unsuitable food and more or less starved develop 'rubber jaw', a severe dysplasia often aggravated by kidney disease. Horses and ponies have developed osteofibrosis when they are fed on cereal concentrates and when the hay ration is kept low. The calcium content of the drinking water is important. Bran has a low calcium and high phosphorus content and horses confined to a dark stable and fed on bran develop osteofibrosis, known as bran disease, or big head, or Miller's disease. Osteofibrosis of horses is common in India, China, and the Philippines, and in the last-named country the calcium content of the feed is so low that calcium has to be added. Bran disease in horses is recognized by the swelling of the head, the curved spine, loss of appetite, and weakness and inability to stand. As the result of the semi-starvation, these animals have a precocious and depraved appetite and will eat anything. It is known as osteophagia, craving for bone, or 'pica' (Groenewald, 1937). It is associated with a deficient phosphorus intake. Groenewald's experiments in two-year-old fillies with a ration of calcium (CaO) 4·5 g. and 54·29 g. P_2O_5, a ratio 1 : 12·06 resulted in clinical symptoms of osteofibrosis. Horses receiving a deficiency in calcium lost weight and became emaciated and poor in condition after eight months and then progress of the disease was more rapid. Blood-calcium determinations are of little value for diagnostic purposes in experimental cases of suspected osteofibrosis. Facial enlargements, when once established by the disease, were not reduced in size by feeding a ration which was supplemented with $CaCO_3$. The pathological changes in the bone resulting from the withdrawal of calcium and phosphorus vary according to the age, mode of life, and species of animal. There is no evidence that these bone lesions in animals are produced by any endocrinological disturbance. If these horses are fed on good pasture grass *early*,

before the loss of appetite and before the disease is well established, they recover.

Infants and children under the age of 6 years with dysplasia of the jaws—so-called cherubism—or early leontiasis ossea improve if given daily doses of cod-liver or halibut-liver oil (vitamin D), additional milk (easily assimilated calcium), and carefully regulated sunlight treatment. The calcium must be in a form easily assimilated such as milk, which has a higher calcium and phosphorus content than blood. It is essential that this diet and any treatment should be commenced early, and before ossification begins. Kidney disease and enteritis produce a rapid deterioration, hence it is desirable to investigate the function of the kidneys. Progress is observed by radiography before and six months after treatment. Biopsy of the bone with strict asepsis when practicable is valuable. Children suffering from dysplasia of the jaws are very few and one clinician does not see a sufficient number of cases to draw conclusions. Moreover, owing to the second world war it has been impossible to follow up these cases.

The osteodystrophies require further investigation from a biochemical and pathological point of view. Frequent biopsies and radiographs of the bone are important. Experimental research on the feeding and diet of animals is difficult. Spacious and well-equipped animal houses and a special dietetic department are essential for such experiments. The grafting of a fresh piece of bone from a case of Paget's disease into an animal is more than difficult.

SUMMARY

Leontiasis ossea is a focal fibrous dysplasia of the bones of the face and it is identical with Paget's disease, which is a diffuse fibrous dysplasia. Osteodystrophy includes those cases of abnormal development and ossification of bone, such as leontiasis ossea and Paget's disease.

The biochemistry of bone and the part played by calcium and phosphorus in the ossification of bone is discussed.

Experiments on the feeding of goats and horses by eliminating calcium and phosphorus are mentioned.

The treatment of children with dysplasia of the jaws, labelled 'cherubism', is given.

Attention is drawn to the desirability of clinical and experimental research in cases of fibrous dysplasia.

Acknowledgements.—I am indebted to the late Sir Edward Mellanby for useful advice; to Professor Sir Charles Dodds for the chemical examination of a number of specimens of bone at the Courtauld Institute; and to the Ferens Institute, Middlesex Hospital, for the photographs and microscope slides; also Mr. E. Cotkin, of the Royal Veterinary College, for useful information.

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LEIOMYOSARCOMA OF THE COLON

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LEIOMYOSARCOMA of the colon is very rare, only 10 cases of this condition having been reported to date. The first case recorded was that of Scott (1923), and further cases were reported by Neugebauer (1932), Trygstad (1932), Golden and Stout (1941), Ripstein and Flint (1952), and Mackenzie, McDonald, and Waugh (1954).

We feel that in view of the complete absence of any reference to this condition in British literature, the following case is worthy of note.

CASE REPORT

H. D., a retired man aged 72, was admitted to the Manchester Northern Hospital on April 5, 1954. He had passed blood per rectum on two occasions. The first occasion was three months prior to admission, and the second nine weeks later. The motions were normal between these attacks. There were no other bowel symptoms, and no loss of weight. Six months previously he had had a left-sided hemiparesis of gradual onset, and was known to have advanced arteriosclerosis.

The patient was of average height, moderately obese, and had a good colour. Both lungs were clear. There was no cardiac abnormality other than changes consistent with his age and the condition of his arteries. He had moderately advanced arteriosclerosis. His blood-pressure was 190/120 mm. Hg, and his pulse and respiration rates were normal.

FIG. 233.—Barium enema: An opaque mass is outlined extending upwards from the false pelvis into the left lumbar region.

There was a smooth, firm, mobile tumour in the left lumbar region. It was in the line of the descending colon. It was reniform, and measured approximately 4½ in. vertically and 3½ in. horizontally. It was not tender.

The rectum contained fæces which were stained darkly with altered blood.

Barium enema on April 8 showed an obstruction present at about the junction of the descending and pelvic colon. After much palpation a thin streak of barium was forced farther upwards, outlining the medial margin of an opaque mass which extended upwards from

FIG. 234.—Photograph of the specimen. Bisection shows the large cystic cavity, the colon flanking the inner border of the tumour.

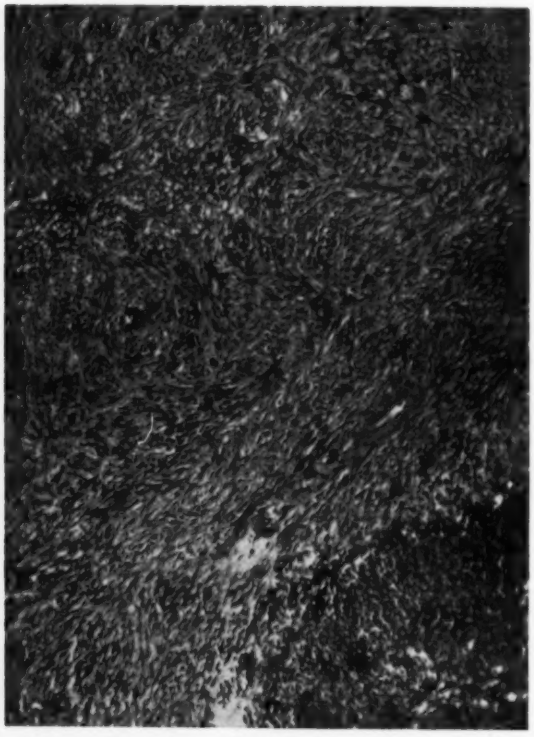

FIG. 235.—Photomicrograph—well-differentiated leiomyomatous structure with typical whorled pattern. (× 75.)

FIG. 236.—Photomicrograph—scattered areas of pleomorphic disordered cells showing nuclear abnormalities and excessive mitotic figures (× 360.)

the false pelvis into the left lumbar region (*Fig.* 233). Two hours after this investigation a motion was passed which contained no overt blood, but four hours later the patient had a brisk hæmorrhage of bright-red blood per rectum. Shock developed, necessitating blood transfusion. The blood-pressure had fallen to 120 systolic by 10 a.m. on the day following the radiographic examination, and remained at this level until twelve hours later, when a further severe hæmorrhage occurred. As his general condition was deteriorating, operation was performed.

AT OPERATION (April 9).—Under general anæsthesia the abdomen was opened through a left paramedian incision. A large, mobile circumscribed mass about 5 in. in diameter was found, involving the descending colon as far down as the commencement of the pelvic loop. There were no obvious secondary deposits. The tumour with 4 in. of bowel above and below was resected with the appropriate glandular field, and end-to-end anastomosis performed. The abdomen was closed with a small drain down to the area of anastomosis.

Following the operation his condition remained fair. Unfortunately cardiac embarrassment supervened, and he died two days later.

THE SPECIMEN (*Fig.* 234).—The specimen consisted of a length of colon to which was attached a smooth reniform tumour, which measured 10·5 cm. in length, 9 cm. in breadth, 8·5 cm. in thickness. The surface of the tumour was lobulated, and completely covered with peritoneum, through which could be seen numerous minute branching vessels. The colon extended for 6 cm. above the tumour and for 5 cm. below it. These portions of the colon showed no abnormality. The portion of the colon in relation to the tumour was flattened, the lumen being reduced to a long anteroposterior slit. Here the mucosa of the bowel was smooth, but showed two shallow ulcers overlying the tumour. One was rounded, 2 cm. in diameter, near the upper extremity of the growth; and the other was linear, 2 cm. in length and 0·5 cm. in breadth, and related to the middle portion of the neoplasm.

Bisection of the specimen showed the tumour to have a well-defined capsule containing a large irregular cystic space, which was filled with blood-clot. The wall of the cyst, which was composed of tumour tissue, was irregular in thickness. It reached a maximum of 2·3 cm. on its medial aspect, and diminished to a minimum of 1 mm. on its lateral aspect. There were trabeculations on the inner surface of the wall of the cyst very similar to the trabeculæ carniæ of the heart.

There was no connexion between the tumour cavity and the lumen of the bowel. The related mesentery showed no lymph-gland involvement.

Table I.—DETAILS OF CASES

AUTHOR	SEX AND AGE	DURATION	PRESENTING FEATURE	CLINICAL FEATURES	BARIUM ENEMA	FINDINGS	TREATMENT	PROGNOSIS
Neugebauer, 1932	F., 48	?	Pain on walking, working	Abdominal mass tender, fixed, size of fist	No conclusive picture	Mobile tumour of cæcum	Resection	1 year good
Trygstad, 1932	M., 19	1 mth.	Pain, severe left-sided abdominal, nausea and vomiting	Abdominal mass left lumbar region, size and shape of kidney, slightly tender, freely movable	Not noted	Tumour ileocæcal junction, with intussusception	Local resection of cæcum. Ileum into ascending colon end-to-side	Not noted
Golden and Stout, 1941	M., 78	6 mths.	1. Pain, slight intestinal hæmorrhage 2. Abdominal mass	Abdominal mass		Sigmoid	Resected	1 year good
Ripstein and Flint, 1952	M., 56		Pain, vague abdominal. Flat. Constipation	Abdominal mass. Marked tenderness right upper quadrant	Filling defect ascending colon	Firm, irregular nodules, mass in cæcum. Intussusception as far as hepatic flexure	Resected	Died 2¼ years after operation. No P.M. Cause not stated
McVerry and Levene, 1956	M., 72	3 mths.	Blood per rectum	Abdominal mass, mobile, smooth, firm, left lumbar region, reniform	Obstruction to flow of barium. Tumour outlined	Mobile tumour of lower end descending colon	Resected	Died two days after operation cardiac failure

HISTOLOGY.—The tumour consisted partly of tissue showing a well-differentiated leiomyomatous structure, with a typical compact whorled pattern (*Fig.* 235). There were scattered areas of pleomorphic disordered cells showing nuclear abnormalities and excessive mitotic figures (*Fig.* 236).

POST-MORTEM FINDINGS.—The lungs showed a patchy bronchopneumonia. The coronary arteries were calcified and the myocardium showed patchy fibrosis. There was generalized arteriosclerosis. The entire tumour appeared to have been removed, and there was no evidence of spread in the abdominal cavity or elsewhere. The anastomosis was sound.

DISCUSSION

Of the 10 cases of leiomyosarcoma of the colon previously reported, we have been able to acquire accurate data on 4 cases. The salient features of these as well as those of the present case are tabulated above (*Table I*). Mackenzie and others (1954), in an analysis of smooth-muscle tumours of the colon, include all such tumours reported to date, both

benign and malignant. As no specific details are given, we are unable to include their cases in our discussion.

From this small series of cases it would appear that leiomyosarcoma of the colon may occur in either sex, from the young adult to the aged. The duration of symptoms appears to be comparatively short. The commonest presenting features were abdominal pain, which occurred in 3 out of 5 cases, and hæmorrhage, which occurred in 2 out of 5 cases. The clinical finding, which was common to all cases, was the presence of an abdominal mass. In 3 of the 5 cases the tumour was tender. Mobility was a notable feature in 2 cases, but both these cases were of short duration. Neugebauer (1932) described the mass as being fixed, but at operation it was found to be ' greatly ' mobile. The known duration of this case was not noted. In the case described by Ripstein and Flint (1952), the clinical picture was complicated by the presence of an intussusception which had progressed as far as the hepatic flexure. Though the presence or absence of mobility was not commented upon, this complication may possibly account for its absence. The longer duration in this case may have had some bearing on this, although it is fair to assume that a cæcal tumour which progresses to the hepatic flexure is mobile. Golden and Stout (1941) do not mention this feature ; thus it is possible that in all these cases the tumour was mobile.

In the 2 cases in which barium enema was performed, a positive finding was reported. Neugebauer (1932) stated that a radiograph of the bowel showed no conclusive picture in his case.

Table II, from Mackenzie and others (1954), shows the site of origin of 10 cases previously reported. It is noteworthy that the present case is the first reported of a leiomyosarcoma of the descending colon.

Table II.— Site Incidence of 10 Cases

Cæcum	3
Ascending colon	1
Transverse colon	3
Descending colon	0
Sigmoid colon	3

In 5 cases reviewed no treatment other than resection was carried out. Two cases were reported as being well one year following the operation. One case died two years and three months after operation, but as no post-mortem was performed the cause of death was unknown.

Diagnosis.—The presence of a lump in the line of the colon, associated with intestinal symptoms, more or less confines the differential diagnosis to that of other benign and malignant growths affecting the colon. From the clinical standpoint it may be impossible to differentiate a leiomyosarcoma from a benign tumour, such as a myoma or a lipoma. These benign growths may cause pain and bleeding as a result of intussusception. But where a mass is associated with melæna, the most likely diagnosis is that of a carcinoma. Until the pathologist's report emerged, the diagnosis in our case was regarded as that of a carcinoma of the descending colon. It is doubtful if any one fact in this case would have led to the pre-operative diagnosis of a leiomyosarcoma of the colon ; but a correlation of all the features present may have suggested it. The history of bleeding alone, without any accompanying mucus or diarrhœa, is unusual in carcinoma. It was noted when the patient was first seen that it was unusual to find a carcinoma of such a size in this site remaining so smooth and mobile. Although it felt firm it was not hard. The barium enema, apart from demonstrating the site of the obstruction to the upward flow of barium, delineated beautifully the eccentric position of the growth in relation to the lumen of the bowel. It is amazing that such a picture could be obtained in the absence of signs of intestinal obstruction. Also, as a result of the manipulation performed during this investigation in the attempt to force the barium past the block, hæmorrhage followed, which finally necessitated a direct attack on the growth to arrest it.

CONCLUSIONS

1. A case of leiomyosarcoma of the descending colon is presented. The unusual features worthy of note are :—

a. This is the first recorded case arising from this site.

b. Bleeding following barium enema necessitating resection to control it.

2. The differential diagnosis has been discussed. These growths are most likely to be diagnosed as carcinomata.

3. If treated along the same lines as carcinoma, the resection is adequate. Hence, accuracy in clinical diagnosis is academic, and may remain as a personal triumph to the clinician concerned.

Acknowledgements.—We are grateful to Mr. H. A. Haxton for his advice and for permission to publish this case which was treated under his care ; to Dr. E. W. Lillie for his help and criticism ; to Dr. Duran Jorda and Miss J. Perry for the photography ; and to Mrs. Michaels for her translation from the German.

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THREE UNUSUAL CASES OF HYPERPARATHYROIDISM

By D. J. M. CRAWFORD, J. STEFANELLI, and A. F. ALVAREZ

THE C. S. WILLIAMS CLINIC, TRAIL, BRITISH COLUMBIA

Case reports of hyperparathyroidism nowadays appear with such regularity that the condition can no longer be considered as a rarity. The occurrence of three such cases within the space of one year, in a relatively small community, may, perhaps, lead one to think that the incidence might be greater than is generally accepted. The presentation of three further cases of hyperparathyroidism is excused on the grounds that each one of them presented some unusual feature. In the first case the age incidence is the second earliest found in a relatively wide survey of the literature ; in the second case the close association of recurrent hyperparathyroidism and recurrent peptic ulceration is striking ; in the last case there is strong histological evidence of malignancy, carcinoma of the parathyroid gland being an extremely rare condition.

CASE REPORTS

Case 1.—The patient, a 3-year-old boy, was referred for consultation on Nov. 12, 1953. The mother stated that he had started to limp one year previously. He was a well-built, sturdy little boy with a cherubic face. On walking there was a moderate limp of the left lower extremity, which seemed to originate in the corresponding hip. No generalized or local abnormality could be made out after a very thorough clinical examination. Radiographic examination of the pelvis (*Fig.* 237) showed a well-defined, circular area of rarefaction in the trochanteric region of the left femur ; it was interpreted as representing a simple bone cyst. The advisability of suitable investigation and surgical exploration was explained and admission to hospital arranged.

The following investigations were made : Hb 94 per cent, W.B.C. 5,500 (œsins. 2 per cent, stabs. 9 per cent, segs. 40 per cent, lymphs. 45 per cent, monos. 4 per cent), basal sedimentation rate 1 mm./hr., serum-phosphorus 2·1 mg. per cent, calcium 11·1 mg. per cent.

As a result of these findings it was assumed that the lesion present was, indeed, that of a simple bone cyst. Operation was undertaken by one of us (A. F. A.) on Nov. 24. The cortex over the greater trochanter was obviously thinned, and it was smooth and dark in colour. The cortex was easily removed, revealing an underlying cystic cavity with even walls, containing a little loose cancellous bone. The cavity was curetted and filled with bone chips, which were obtained from the corresponding iliac crest through a separate incision. The limb was immobilized in a Thomas's splint with skin traction. The post-operative progress was uneventful.

The histological report on the tissue excised was as follows :—

Gross description : Numerous portions of rather granular-appearing osseous tissue, aggregating to a volume of almost 2 c.c. The entire specimen will be embedded and will be decalcified prior to sectioning.

Microscopic : Sections through the small portion of osseous tissue, after decalcification, show these to consist of a number of portions of osseous tissue apparently undergoing some slight degenerative change and they are interspersed with a moderately compact, slightly œdematous fibrous connective tissue in which a little inflammatory reaction is present and, in an occasional instance, rather cyst-like formations are noted in this fibrous tissue. About the periphery there is some rather denser connective tissue in which some lymphocytic infiltration is present and a few scattered osteoclastic cells are noted, here and there, throughout this interosseous fibrous tissue. I believe that this is in the nature of an osteitis fibrosa cystica.

Figs. 238, 239 show low- and high-power microscopic views, respectively, of the tissue examined.

The histological diagnosis was regarded with some reservation, but none the less a reinvestigation of the pertinent blood chemistry was ordered. The results

Fig. 237.—*Case* 1. Cyst of left trochanteric region.

reported on Dec. 7 were : alkaline phosphatase 4 Bodanski units, serum-phosphorus 4 mg. per cent, serum-calcium 11 mg. per cent.

The original diagnosis of benign cyst being thus apparently confirmed, the patient's limb was immobilized in a plaster spica, and the patient discharged one week later.

The spica was removed on Feb. 4, 1954. The general condition of the patient was excellent and the mother was very pleased with his progress. The radiographic appearances at the time showed satisfactory filling of the cystic cavity. The urine was still negative and there was no substantial change in the hæmoglobin level or in the white-cell count. The serum levels were : alkaline phosphatase 2 B.U., phosphorus 2·5 mg. per cent, and calcium, 12·4 mg. per cent. The last figure caused some alarm, but, in view of the fact that the patient was symptom-free and thriving, it was deemed that there was no cause for further interference at the time ; it was decided to maintain the out-patient supervision with renewed care.

When the child was seen again in April, the mother stated that he had recently started to limp anew. Once more there was no abnormality on examination, barring

the presence of the two operative scars. A radiograph of the pelvis showed satisfactory progress in the repair of the cystic cavity. Films were taken of the long bones of the extremities, and although these did not reveal any further cysts, an abnormal degree of rarefaction seemed to be present. The possibility of hyperparathyroidism was again seriously considered and further investigation advised.

The urine was still free from any abnormality. The urea concentration test showed urea levels in the urine of 2·2, 1·2, and 1·65 g. per cent. Blood-count: R.B.C.s 4,430·000; W.B.C.s 5,500; Hb 76 per cent, slight anisocytosis and hypochromia were present. Blood-urea 40 mg. per cent. Serum levels: phosphorus 4 mg. per cent, serum-calcium 13·2 mg. per cent, and a few days later, alkaline phosphatase 3·2 B.U., phosphorus 4·1 mg. per cent, calcium 14·2 mg. per cent. The Sulkowitch test showed the presence of only a trace of calcium in several twenty-four-hour specimens of urine. At the conclusion of these investigations it was felt that the presence of hyperparathyroidism was now too likely a possibility to be ignored any longer, and surgical exploration was advised.

Operation (A. F. A.) was carried out on May 4. A thorough exposure of the entire thyroid gland was made, special care being exerted in displaying the posterior surfaces of each lobe and both recurrent laryngeal nerves. The only evidence of parathyroid tissue encountered corresponded to a small pinkish structure, 0·5 × 0·3 cm. in diameter, in the position characteristic of the left superior gland; no other parathyroid tissue, normal or otherwise, was found in the neck, or, by palpation, in the anterior mediastinum. The tissue referred to was removed together with another rounded structure which resembled an enlarged lymph-gland.

The histological report received from Dr. H. Pitts was as follows:—

"Gross description: The specimen comes in two portions, the first being a small ovoid nodule of presumed parathyroid origin measuring 0·5 × 0·3 cm. The second portion is a quantity of fibro-fatty tissue containing firmer material in the central portion, the whole aggregating to a mass 0·8 × 0·4 cm.

"Microscopic: Sections through the material from the neck show one of the specimens to consist of typical parathyroid gland and the other to consist of lymphoid tissue. The parathyroid gland tissue is composed of small cells arranged in indefinite alveolar aggregations and the cells generally possess small, very regular-appearing nuclei. The gland is quite well vascularized throughout. It is larger than can be considered commensurate with the normal and its histology suggests a benign adenoma. Sections through the lymphoid tissue show an inflammatory hyperplasia of a non-specific type. There is no evidence of any tumour or granulomatous involvement.

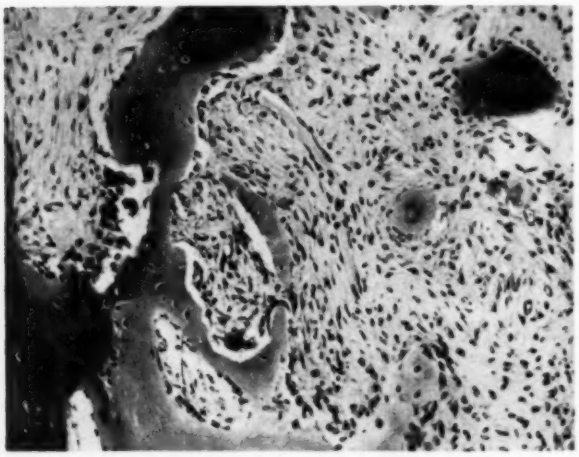

FIG. 238.—*Case* 1. Low-power view of tissue from bone cyst. (× 60.)

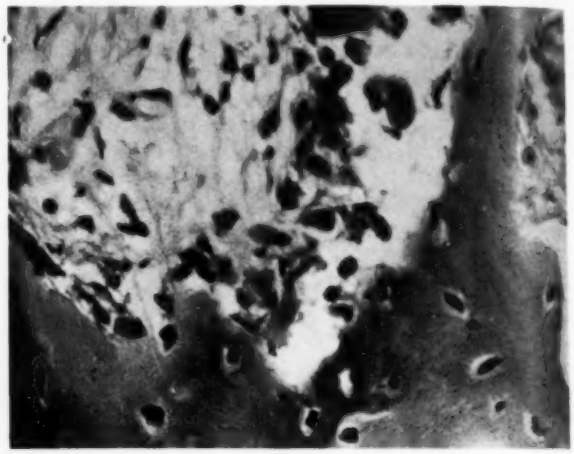

FIG. 239.—*Case* 1. High-power view of tissue from bone cyst. (× 300.)

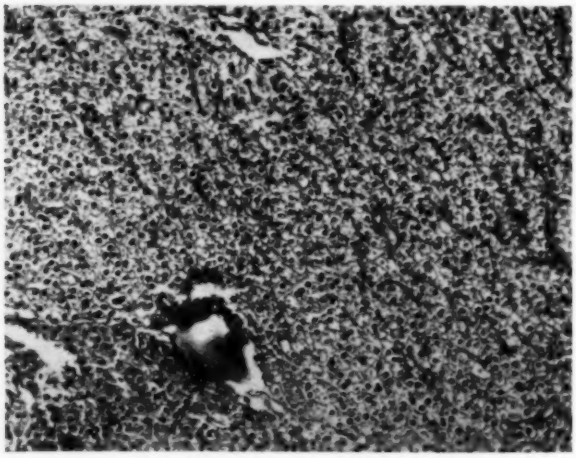

FIG. 240.—*Case* 1. Low-power view of parathyroid adenoma. (× 60.)

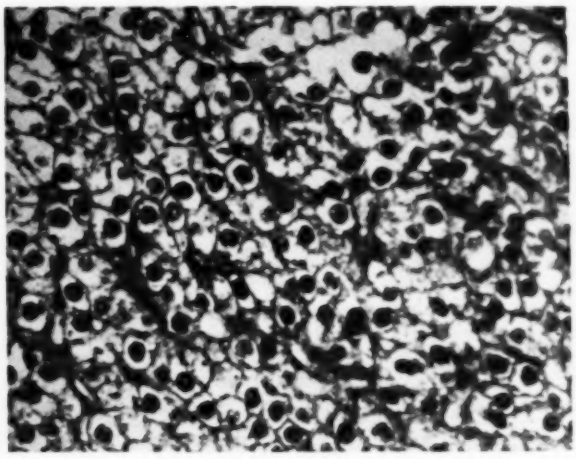

FIG. 241.—*Case* 1. High-power view of parathyroid adenoma. (× 300.)

"Diagnosis: Parathyroid adenoma."

Figs. 240, 241 show low- and high-power microscopical views of the adenoma.

The post-operative progress was uneventful. Six days after operation the pertinent serum levels were: alkaline phosphatase 4·0 B.U., phosphorus 3·8 mg. per cent, calcium 12 mg. per cent; nine days after operation they were 5·2 B.U., 3·1 mg. per cent and 11·9 mg. per cent respectively.

FIG. 242.—*Case* 1. Healing bone cyst in left trochanteric region.

Since then the child has been in excellent condition. The limp has disappeared and he has been developing as a healthy boy of a similar age, and it might be said that he is above average. In January, 1955, he was readmitted to hospital in order that a careful review of his progress could be made. Radiographic studies of the long bones showed decreased decalcification; *Fig.* 242 shows the progress made in healing of the previous cyst. A blood-count was reported as follows: Hb 12 g. or 76 per cent, W.B.C.s 7,250 (œsins. 1 per cent, segs. 61 per cent, lymphs. 32 per cent, monos. 6 per cent), the morphology being regular. The urine showed, for the first time, the presence of albumin, varying from +1 to +2. Blood and serum levels: urea 40 mg. per cent, proteins 6·2 g. per cent (albumin 4·6, globulin 1·8), serum-chlorides 585 mg. per cent, alkaline phosphatase 4·0 B.U., phosphorus 3·3 mg. per cent, calcium 11·7 mg. per cent. The last three results were repeated a few days later, giving values of 12·5 K.A. units, 5·8 mg. per cent and 10·3 mg. per cent respectively. An intravenous pyelogram failed to reveal any abnormality, there being no delay in dye secretion.

We are a little concerned at the recent appearance of albumin in the urine, although the absence of casts and other abnormalities, together with the normal pyelogram, militate against the presence of any serious renal lesion. This aspect of the case will also be kept under supervision.

Although the patient was first seen at the age of 3 years, it must be assumed that hyperparathyroidism must have been present when the symptoms first appeared, if not earlier. As the limp had been present for one year prior to seeking medical attention, it seems fair to claim the onset to have occurred at least at the age of 2 years. Anspach and Clifton (1939) report a case in which symptoms developed at the age of 3 months. The parents refused permission for operation and although a striking remission of symptoms and signs have followed deep X-ray irradiation, there is no histological proof of the presence of an adenoma. The clinical picture given is so characteristic, however, that any other diagnosis but that of primary hyperparathyroidism is unlikely. With this exception, no other case of hyperparathyroidism due to adenoma has been found to be recorded at such an early age in a fairly wide survey of the literature on the subject.

Case 2.—The patient, a 30-year-old man, first came under the formal attention of the Clinic in April, 1949. He was suffering from pathological fractures involving the right humerus, the right femoral shaft and neck, and the left femoral neck. These are clearly shown in *Figs.* 243, 244, which also demonstrate the presence of extensive bone rarefaction and cystic formation. These, together with the presence of kyphosis, bowing of the femora, and other deformities, were very suggestive of osteitis fibrosa cystica of the generalized type. This was confirmed by the presence of a raised serum-calcium and a lowered serum-phosphorus, 13·8 mg. per cent, and 2·8 mg. per cent, respectively. The Sulkowitch test at the time showed an excess of calcium in the urine to the extent of 1+.

The previous history is of some interest as he was reported to have had some "stomach trouble" in 1943, at the age of 24 years, and pulmonary tuberculosis in 1945, at the age of 26 years.

An exploration of the neck was performed in August, 1949, by the late Dr. M. Basted. At this, two adenoma-like tumours were reported to be present, one in each side of the neck, in the positions characteristic of the inferior parathyroid glands. They were excised. Subsequent histological examination by Dr. H. H. Pitts reported one of these structures to be composed of typical parathyroid tissue; the other was said to be composed of fatty tissue only.

Post-operatively the serum-calcium dropped to 7 mg. per cent. At the time of discharge from hospital, roughly one year after admission, the serum-calcium was 9 mg. per cent, and the serum-phosphorus 4·2 mg. per cent. The various fractures present had been treated by our colleague, Dr. M. E. Krause, and were soundly united.

The patient was readmitted to hospital in October, 1951, and in July, 1952, because of the presence of blood and pus in the urine. The diagnosis made in each instance was that of severe pyelonephritis.

In October, 1952, at the age of 33 years, further readmission was necessitated because of the onset of severe hæmatemesis. At this time a barium meal was performed and reported as negative. The urine showed no abnormality. The serum-calcium was 11·2 mg. per cent and the serum-phosphorus 2 mg. per cent. We were asked to see the case in consultation; in view of the presence of intermittent stomach trouble since the age of 24 years, which was strongly suggestive of peptic ulceration, and the occurrence of hæmatemesis, laparotomy was recommended. Parathyroid function, at the time, was thought to be normal.

Operation was carried out by one of us (A. F. A.) in November, 1952. A lesser-curve ulcer of the stomach, quite high in situation, was found, and a three-quarter resection of the stomach was performed. A Billroth I type of anastomosis was established. Recovery was uneventful.

The patient was well until April, 1953, when he complained of pain in the left hypochondrium passing to the right flank, and also upwards to the chest and to the left shoulder. The pain was more prominent before

meals and was relieved by food. The presence of a stomal ulcer was suspected. The diagnosis was evident in May of the same year, when severe pain in the epigastrium appeared and was followed by two bouts of copious

FIG. 243.—*Case* 2. Pathological fractures of right femoral neck and shaft. Abnormal calcification of vessels in the thighs.

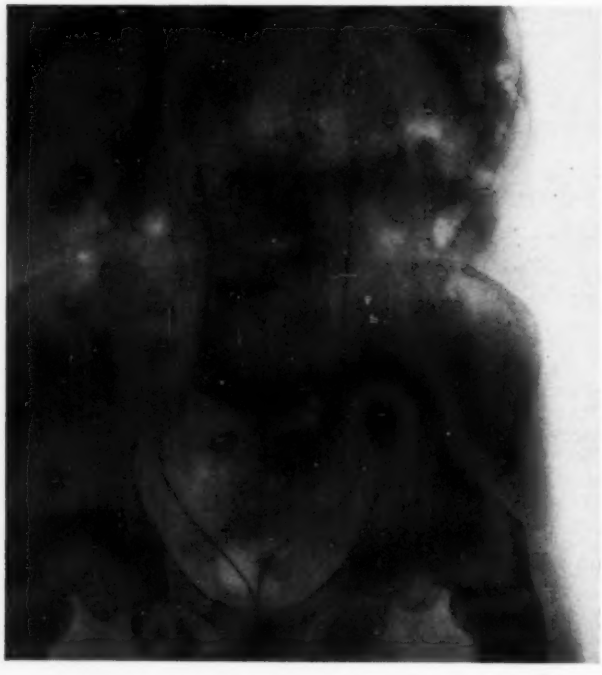

FIG. 245.—*Case* 2. Retrograde pyelogram showing moderate bilateral hydronephrosis. Calcification in the region of the left kidney should be noted.

hæmatemesis. The patient was readmitted to hospital. A fractional test meal revealed a high free and total acid content in the stomach, whilst in the barium meal a duodenal defect was reported. The consternation ensuing from such stomal ulceration led to a complete disregard of the possible persistence of hyperparathyroidism. A further laparotomy was performed (A. F. A. and J. S.) in June, 1953. The presence of a stomal ulcer having been established, a higher resection and a vagotomy were carried out, a Hofmeister-Lake-Finsterer anastomosis being employed this time. Once again recovery was uneventful.

In August, 1953, two months after operation, the patient complained of pain in the right hypochondrium.

FIG. 244.—*Case* 2. Pathological fracture of the surgical neck of the left humerus.

He also stated that he was full after meals and that he felt weak. The patient was seen at frequent intervals thereafter. In March, 1954, we had to admit, loath though we were, that further stomal ulceration was present. A moderate anæmia was present at the same time. Treatment with a 'gastric' diet and alkalis was instituted as an Out-Patient, with only partial success. In November, 1954, the patient was readmitted into hospital for further investigation. As pus and red blood cells were present in the urine a urinary investigation was also deemed to be necessary.

The results of the various analyses performed were as follows: Intravenous pyelogram, very poor filling: retrograde pyelogram (*Fig.* 245), moderate bilateral hydronephrosis; pus cells in the specimen from the left ureter and *Esch. coli* cultured from the bladder urine; blood-urea 30 mg. per cent; urine specific gravity 1·008, a trace of albumin being present; urea concentration test, maximal and minimal concentrations 0·95 and 0·70 per cent respectively. Further radiological studies of the limbs showed the presence of extensive calcification of the vessels and of the soft tissues; the lamina dura normally surrounding the teeth was absent. The extent of the osteoporosis and cystic changes thus revealed was such as to be deemed inconsistent with a return to normal parathyroid function four years previously. Pertinent serum and urinary estimations were made on two occasions and were reported as: alkaline phosphatase, 3·2 and 2·8 B.U., phosphorus, 2·3 and 2·6 mg. per cent, calcium 12·4 and 12·5 mg. per cent, Sulkowitch +3 and +2. The radiological and biochemical evidence was thus strongly in favour of the persistence of hyperparathyroidism. Meanwhile a further barium series was performed and the presence of a stomal ulcer was reported.

The therapeutic problem thus presented was formidable. Unfortunately we were not aware, at the time, of

the close relationship that exists between hyperparathyroidism and peptic ulceration. It was clear, however, that both conditions could not be treated at one and the same time. In view of the presence of severe renal disease it was thought preferable that attention should first be directed to the control of hyperparathyroidism. Besides, should exploration prove to be negative, laparotomy need not be long delayed.

At operation (A. F. A. and J. S.) both lower parathyroid glands were clearly enlarged, each being about 1·5 × 1 cm. in diameter. They were removed without undue difficulty. Dr. H. H. Pitts reported upon the histological appearances (*Fig.* 246) as follows :—

" The first part of this specimen is said to be from the right parathyroid area and consists of a mass of connective tissue and several small nodules, the total measuring 2 cm. in diameter. There are three nodules within this mass, the two smaller measuring 6 mm. in diameter, and the larger one 3 cm. in diameter. The entire specimen will be embedded and labelled ' A '. The second portion from the parathyroid region of the left side consists of a similar mass of connective tissue containing three nodules measuring 1·5 cm. in diameter. One of the nodules is soft and friable and measures 5 mm. in diameter ; the rest of the mass consists of a very firm calcified tissue which includes a node measuring 1 cm. in diameter. The entire specimen will be embedded and labelled ' B '.

" Microscopic : Sections through the lesion labelled ' A ' show a well-encapsulated adenoma composed of parathyroid tissue in which the individual cells are arranged in small aggregations separated one from the other by thin fibrous septæ. The cells contain abundant lipoid and the nuclei are generally relatively small, although some are large and vesicular. There is no apparent mitotic activity. Both alpha and beta cells are identified. Sections through the material labelled ' B ' again show a benign parathyroid adenoma, which, however, appears to have undergone rather extensive interstitial fibrosis and there has been some deposition of calcific material in the peripheral areas. There is no evidence of any malignancy in any of these sections.

" Diagnosis : Parathyroid adenomata, benign."

Post-operatively the patient complained of severe tinglings down the forearms. No carpopedal spasm followed arterial occlusion and the Chovstek sign was negative. Such symptoms were promptly relieved by the administration of calcium gluconate intravenously and a high vitamin D and calcium diet. Several serum and urine analyses were made during this period, viz. :—

Post-operative Days	*Ca*	*P*	*Alkaline Phosphatase*	*Sulkowitch*
1	6·8	2·2	2·4	Negative
4	10	—	—	"
6	8·3	2·9	—	"
14	7·5	—	—	"
17	—	7·6	1·6	"
26	8·5	3	5·2	Trace

The striking result was the sudden cessation of all dyspeptic symptoms from the day of operation. These had been very severe and had persisted right up to the day when surgery was undertaken. A barium meal performed four weeks after operation no longer revealed the presence of a stomal ulcer. The patient has been seen at repeated intervals since then and he remains symptom-free.

It is only recently that we have become aware of the well-documented relationship between hyperparathyroidism and peptic ulceration, and we cannot help feeling a certain elation at having drawn the correct inferences when faced with a problem the solution of which was then unknown to us. We have undertaken certain clinical investigations attempting to correlate the effects of an increased serum-calcium level with hyperchlorhydria, but we are at too early a stage to make any further comment on this subject.

Case 3.—The patient, a 41-year-old female, had been under observation on and off since 1951. She had been complaining of vague pains which were widespread

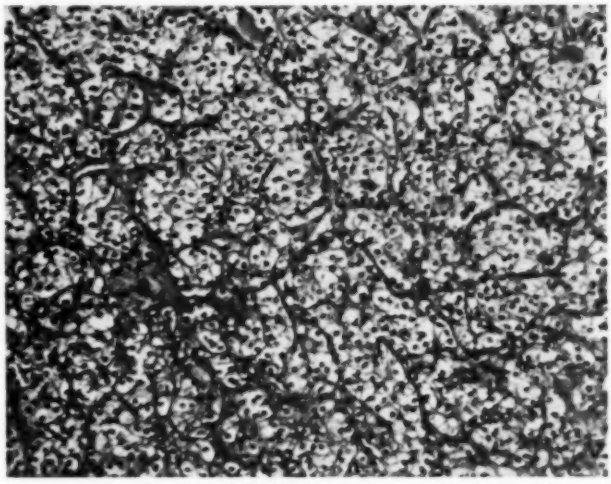

FIG. 246.—*Case* 2. High-power view of parathyroid adenoma.

throughout the body, but most marked in the limbs, lassitude, overweight, and irregularities in the menstrual cycle which were interspaced with phases of amenorrhœa. In 1947 she had undergone a thyroidectomy elsewhere. On July 29, 1954, she presented herself with a swelling in the premaxillary area which was thought to be inflammatory in origin. For the previous three or four weeks she had also been suffering from bilateral episcleritis, for which purpose she had been attending the ophthalmological department. The swelling was incised superficially, and as no pus resulted, she was given intramuscular injections of penicillin. A few days later she was referred to the surgical department as there was no improvement. The patient was admitted to hospital and the administration of penicillin continued at 3-hourly intervals. This was followed by some local improvement, but the patient then began to manifest a raised, ' swinging ' temperature and complained of headache and dizziness. There was no lymph-gland enlargement in the neck, the internal jugular veins appeared to be normal, and there was no œdema of the conjunctivæ. A sample of urine showed a specific gravity of 1·005, both sugar and albumin were absent, and there were 8–10 W.B.C.s per field, as well as numerous epithelial cells. The hæmoglobin was 15·3 g. per cent or 90 per cent, and the white blood-cell count 11,650 (stabs. 2 per cent, segs. 74 per cent, lymphs. 21 per cent, monos. 3 per cent). One week after admission streptomycin (0·5 g. 6-hourly) was given and penicillin administration discontinued shortly thereafter. One week later the temperature was normal and there was some decrease in the size of the swelling. The administration of antibiotics then ceased. Dizziness persisted and the patient also complained of epigastric pain. A further clinical examination at the time showed that there was poor air entry at the base of the right lung and numerous râles were to be heard at this site. The abdomen was slightly, but diffusely distended, and on palpation a doughy sensation was imparted to the fingers ; there was well-marked tenderness in the left hypochondrium. A radiograph of the chest showed some abnormal shadowing passing from the right

hilum towards the corresponding cardiophrenic angle; it was interpreted as representing an area of 'pneumonitis'. A radiograph of the maxilla showed the presence of a swelling which apparently was restricted to the soft tissues, there being no obvious bony involvement. On Aug. 21 the patient began to have severe and repeated bouts of epistaxis; the hæmoglobin level dropped to 8·7 g. (51 per cent) and a blood transfusion was required. The white-cell count meanwhile had risen steadily to

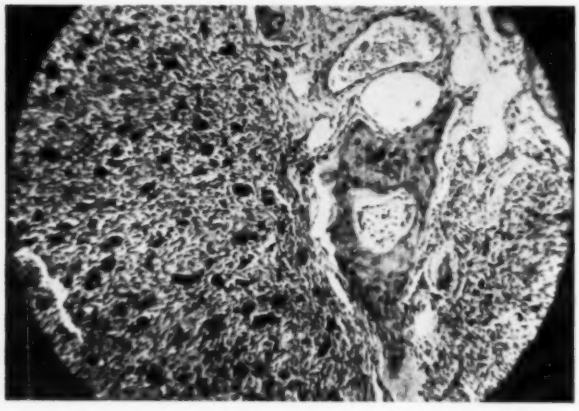

FIG. 247.—*Case* 3. Low-power view of osteoclastoma of maxilla.

25,560, of which 81 per cent was made up by polymorphs. The bleeding time was 1 minute, but the venous clotting time was raised to 14 minutes; the prothrombin index was 14 per cent in spite of repeated checking. The administration of large doses of vitamin K was commenced on Aug. 21, and continued until on Sept. 2 the prothrombin index had been restored to near normal levels. A bromsulphalein test yielded a normal result, apparently eliminating the liver as a factor in this respect.

It had become quite clear for some time that the maxillary swelling corresponded to some form of neoplasm with superimposed œdema and infection. The latter complications having been overcome and the general condition being much improved, surgical exploration of the tumour was recommended. This was carried out (A. F. A. and J. S.) on Sept. 3. A transverse incision was made with diathermy in the superior gingivo-labial fold. With the help of a periosteal elevator the tumour was found to shell out readily, leaving behind a smooth, regular cavity in the underlying maxilla. There was no invasion or penetration of the nasal or antral cavities. The wound was packed with 'Vaseline' gauze. Recovery was uneventful and the maxillary defect remained clean and began to fill up rapidly.

Sections prepared from the tumour (*Fig.* 247) were reported on by Dr. H. H. Pitts as follows:—

"Gross description: A somewhat lobulated, firm mass about the size of a small walnut. On one surface it is covered by thickened whitish membrane, and beneath this, on section, is seen a well-circumscribed, firm, greyish-brown tumour mass which has apparently a few fine fibrils of osseous or calcareous materials scattered throughout its substance.

"Microscopic: Sections through the lesion show very dense fibrous stroma whose cytoplasmic elements are arranged in interlacing fashion, and throughout which there is an occasional collection of mature fat. The nuclei are generally vesicular in type, but some of them are smaller and hyperchromatic, and spindle-shaped in outline. The tumour is interspersed by innumerably larger and smaller giant-cell formations, many of them containing up to a hundred nuclei. Here and there, occasional fragments of osseous tissue are found, and there appears to be abundant osteolyte material in the periphery. Histologically this is a giant-cell tumour, but since it arises from the maxilla is probably best classified as a giant-cell epulis. In any event, it does not appear to be malignant histologically.

"Diagnosis: Benign giant-cell epulis of maxilla."

The term 'benign giant-cell epulis' is nowadays looked upon with suspicion. Some deny the existence of such a primary lesion, considering it to be but a

FIG. 248.—*Case* 3. Radiograph of tibiæ showing osteoporosis and areas of decreased densities resembling small cysts.

localized manifestation of some other generalized lesion in the body. With this in mind, and in view of the patient's widespread complaints and abnormalities, as yet unexplained, it was asked that the slides be reviewed. The pathologist's opinion remained unchanged, but the suggestion was made that if bone were involved in the lesion, then the tumour might be considered to be an osteoclastoma.

The diagnosis of osteoclastoma having been made, the possibility of hyperparathyroidism suggested itself at once, and a full investigation was made to elucidate this point. The pertinent serum estimations, made on several occasions were as follows:—

	Sept. 23	*Sept.* 30	*Oct.* 4	*Oct.* 15	
Alkaline phosphatase	7·2	1·6	4·4	1	B.U.
Phosphorus	2·1	2·8	2·8	2·1	mg. per cent
Calcium	11·1	12·2	11·1	11·8	mg. per cent

The Sulkowitch test for calcium in the urine persistently gave raised values, in the range of +3 and +4. X-ray pictures of the long bones (*Fig.* 248) and of the skull showed the presence of well-marked osteoporosis with scattered areas of decreased density which resembled small cysts. The lamina dura of the teeth was clearly absent. There was also well-marked calcification of both the blood-vessels and some of the soft tissues in the limbs. The radiological appearance *per se* could not be said to be characteristic of hyperparathyroidism (excepting perhaps the disappearance of the lamina dura), for similar changes may also be encountered in multiple myelomata, and in certain instances of bone metastases. There was no Bence-Jones protein in the urine, the sternal marrow was essentially normal, and, in any case, the overall picture was strongly in favour of hyperparathyroidism.

Other investigations were made: Plasma-proteins 6·7 g. per cent (albumin 3·9, globulin 2·8); blood-urea 28 mg. per cent. Urine concentration-dilution test: maximal concentration, specific gravity 1·010; minimal 1·006. Urea concentration test: maximal concentration of urea 0·8 g. per cent; minimal 0·4 g. per cent. An intravenous pyelogram showed gross impairment in the excretion of dye.

FIG. 249.—*Case* 3. Low-power view of parathyroid tissue removed at operation.

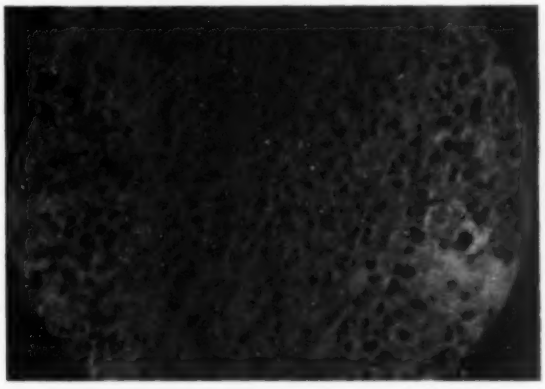

FIG. 250.—*Case* 3. High-power view of parathyroid tissue removed at operation.

Surgical exploration of the neck was undertaken on Oct. 19 (A. F. A. and J. S.). Considerable fibrosis was present in all planes (a legacy of the previously performed thyroidectomy) which rendered the dissection extremely difficult. The four typical sites of the parathyroid glands were none the less clearly displayed. Below the lower pole of the right lobe of the thyroid there was a small pale structure about 1·5 cm. in diameter which was separate from the gland and which carried a blood-supply of its own. This structure was carefully dissected out and removed, together with some pale friable tissue which lay in close relationship with it, again distinct from the thyroid itself. No other parathyroid-like tissue was to be detected in the neck or in the anterior mediastinum. The wound was closed with drainage.

Dr. H. H. Pitts reported on the tissues removed (*Figs.* 249, 250) as follows:—

"Gross description: An irregular portion of very friable, very granular, and finely papillary yellowish-grey tissue, only moderately well encapsulated, and measuring almost 2 × 1 cm. The entire specimen will be embedded. With the specimen is a spherical, well-encapsulated thyroid adenoma measuring 1·5 cm. in diameter, and on section presenting a relatively uniform yellowish-pink cut surface, in which some hæmorrhagic extravasation has occurred. The capsule is more than 1 mm. in thickness.

"Microscopic: A great many sections were taken through the material presumed to be parathyroid tissue and they show small alveolar aggregations of clear cells and other cells containing cytoplasmic granules. The nuclei of the cells are fairly regular in some places, but in others they are extremely irregular, most of them being almost giant-like and mitotic figures are relatively numerous. Throughout there are enormous numbers of small vascular channels and the normal pattern of the parathyroid gland in these areas is moderately well preserved. The capsule of the parathyroid adenoma is well retained in most places, but here and there there appears to be some capsular invasion by irregular aggregation of cells. In one of the sections there is invasion of the walls of some of the peripherally placed blood-vessels. A diagnosis of malignancy is made on purely histological grounds and is made with some trepidation in view of the fact that the lesion is apparently extremely rare.

"Sections through the thyroid adenoma received separately show some slight hyperplasia of the lining follicle cells but there is no suggestion here of malignancy and there is no metastasis from the parathyroid adenoma to the thyroid gland.

"Diagnosis: Malignant adenoma of parathyroid."

The post-operative progress of the patient was uneventful. Two pints of blood were given by transfusion on Oct. 25 as the hæmoglobin level had dropped to 10·8 g. (64 per cent). Post-operative serum estimations were made on several occasions, viz.:—

	Oct. 22	*Nov.* 1	*Nov.* 8	
Alkaline phosphatase	6·4	6·2	4·9	B.U.
Phosphorus	—	3·9	3·2	mg. per cent
Calcium	11·4	10·7	10·4	mg. per cent

The Sulkowitch test remained strongly positive.

In view of the distinct possibility of malignancy a course of deep X-ray therapy was commenced whilst the patient was still in hospital; it was continued subsequently as an Out-Patient service. The patient was discharged on Nov. 9 in a much improved condition.

Readmission to hospital was necessitated on Dec. 14, 1954. Since returning home the patient had been very weak, had no appetite, and was said to have suffered from repeated colds. Amenorrhœa had been present since July of that year. Attendance to the Deep X-ray Department had been discontinued as the patient did not feel well enough; up to that time 3,500 r had been given. On examination one was struck by the intense pallor and the severe emaciation present. A blood-count was taken with the following results: R.B.C.s 2,760,000 (marked anisocytosis present), W.B.C.s 6·950 (œsins. 6 per cent, stabs. 1 per cent, segs. 74 per cent, lymphs. 15 per cent, monos. 4 per cent). A sternal marrow examination was reported as being essentially normal.

Four pints of blood were transfused and were followed by some improvement; one week later the red blood-cell count was raised to 3,890,000 and the hæmoglobin to 13·9 g. per cent (82 per cent). The fæces were tested for occult blood, but none was present. Urine: specific gravity 1·008, albumin +1, no sugar or urobilinogen present. Fractional test-meal: achlorhydria. Serum-cholesterol 165 mg. per cent. Non-protein nitrogen 45 mg. per cent. The Thorn test and the Kepler water test failed to reveal any evidence of adrenal insufficiency. A spinal puncture was made and the cerebrospinal fluid examined: sugar 74 mg. per cent, proteins 20 mg. per cent, no increase in globulin content, chlorides 696 mg. per cent, Wassermann negative, colloidal gold curve 0110000000. Radiological examination of the bones did not suggest any marked change from the previous appearances, although rarefaction seemed to be a little less marked. Barium meal: small sliding hiatus hernia

present; generalized hypermobility of the upper alimentary tract. Intravenous pyelogram: fragmentary filling of the renal pelves, but no definite delay in dye excretion.

	Dec. 20	*Dec.* 31	*Jan.* 17	*Feb.* 7	
Alkaline phosphatase	4	7·5	5·0	8·0	B.U.
Phosphorus	2·2	3·5	3·5	2·2	mg. per cent
Calcium	10·5	10·5	8·8	9·5	mg. per cent

It was thought that the process of hyperparathyroidism had been arrested, but now we were confronted with a severe degree of anæmia and an advanced stage of nephritis. The nature of the blood deficiency was not clear; it could not be ascribed to further external, or internal hæmorrhage, nor was there any evidence of hæmolysis. In view of the fact that a previously performed subtotal thyroidectomy had been followed by a considerable radiation dosage to the same area the possibility of hypothyroidism suggested itself and was compatible with the anorexia and inertia present. The serum-cholesterol level, however, was against such a belief. None the less thyroid extract was administered, at first gr. 4 daily, which was subsequently reduced to gr. 2 daily; some improvement resulted. Although the patient was far from well she insisted upon her discharge on March 6, 1955. A blood-count taken two weeks previously showed a hæmoglobin level of 12·2 g. (72 per cent), and 7,700 W.B.C.s (stabs. 1 per cent, segs. 85 per cent, lymphs. 12 per cent, monos. 2 per cent).

One week later the patient was readmitted in a moribund condition. The blood-count at the time was as follows: R.B.C.s 1,500,000, Hb 5·1 g. per cent (30 per cent). Basal sedimentation rate: 88 mm./hour. Urine: specific gravity 1,005, albumin +2, sugar negative, W.B.C.s 1–3 per field. A radiograph of the chest showed scattered areas of opacity in the lower lobe of the right lung, which were thought to be inflammatory in origin, but the possibility of metastases could not be ruled out. The patient died on March 23. A failure in bone-marrow function was thought to be an important factor in bringing this about.

A post-mortem examination was conducted by Dr. L. Duggan, newly appointed pathologist to the Trail-Tadanac Hospital. The findings are of interest and are reported in full:—

"External examination: An emaciated, slightly jaundiced female. Pupils equal and in the cadaveric position. An old transverse scar was present on the neck approximately 1½ in. above the clavicle.

"Internal examination: Larynx, trachea, and bronchi showed mild chronic inflammation. Lungs: both lower lobes showed extensive firm, grey, consolidated areas involving approximately one-third to one-half of each lobe. The cut surfaces of these areas were firm and gritty. Other areas of the lungs showed a bronchopneumonia. Urogenital system: the kidneys were contracted and extremely scarred. The capsules were adherent. The cut surfaces were slightly gritty. Uterus, bladder, and genitalia normal. Gastro-intestinal system: Œsophagus, stomach, intestines, gall-bladder and pancreas normal. Liver normal in size; the cut surface showed extensive fatty degeneration. There was no evidence of metastases in the liver. Cardiovascular system: The heart was slightly enlarged and was distended by fluid blood and blood-clot. The wall of the left auricle was studded with firm calcareous plaques, extending down to the root of the mitral valve. The valves were normal. There was moderate calcification of both coronary arteries, but the lumina were intact. The aorta and other great vessels appeared normal. Hæmopoietic and endocrine systems: The spleen was of normal size and pattern. The thyroid site showed extreme fibrosis, but some thyroid tissue appeared to be present. The adrenals appeared to be normal. Musculo-osseous system: Fusiform swellings were present on the right 4th, 5th and 6th ribs and on the left 4th rib, in each case approximately halfway between the anterior and posterior articulations. The bones of the vault of the skull showed two areas of thinning each approximately 3 in. behind the anterior end of the sagittal suture and approximately 1 in. on either side. These areas showed smooth hollowing of the bone from within the skull and each area was approximately ½ in. in diameter. There was no evidence of secondary carcinomatous deposits. The bodies of the sacral vertebræ were normal. Central nervous system: Brain and meninges normal.

Histology: Lungs: (1) Bronchopneumonia; (2) Linear calcification following the outlines of the walls of the alveoli, which are lined by concentric fibrous tissue laminæ reducing the alveolar cavities to small central

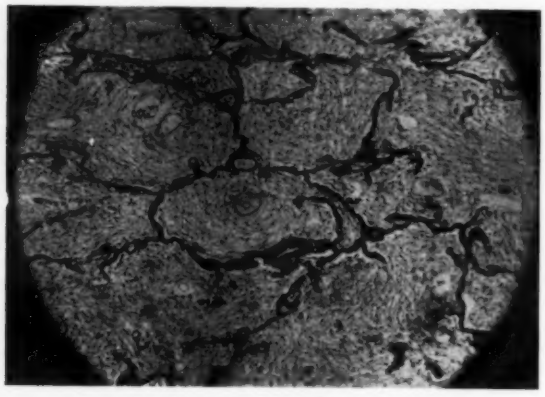

FIG. 251.—*Case* 3. Calcification in lung tissue.

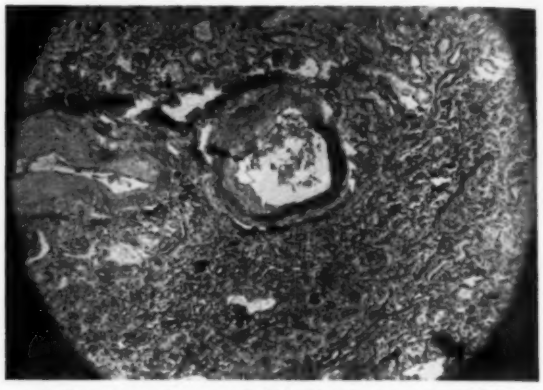

FIG. 252.—*Case* 3. Calcification in a renal arteriole.

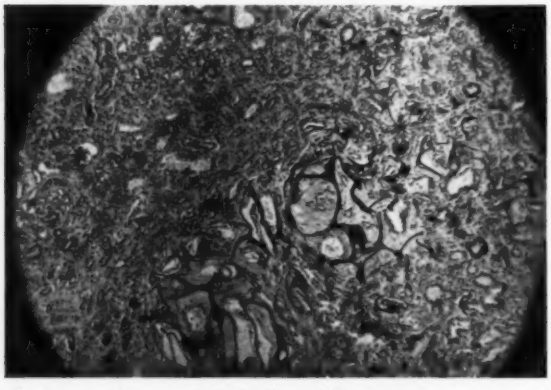

FIG. 253.—*Case* 3. Calcification within the kidney substance (nephrocalcinosis).

circular spaces. Kidneys: Nephrocalcinosis. Extensive destruction of glomeruli and tubes. Interstitial fibrosis, extensive calcification of arterioles and some tubular calcification. Liver: Fatty degeneration. Thyroid: Sections of tissue adjacent to the thyroid show some groups of epithelial cells arranged in trabeculæ and rudimentary acini. In most places these cells are uniform in morphology. No mitoses were seen. There is no sign of a capsule, but this tissue has been subjected to trauma. In one small area there is considerable cellular pleomorphism reminiscent of that seen in the original biopsy. Apart from this we have at the moment no other evidence that the tumour is malignant. There may well have been changes in the original cellular morphology due to radiotherapy. Ribs: Sections of the fusiform swellings showed healed fractures with no evidence of carcinoma deposits.

Cause of death: (1) Bronchopneumonia; (2) Uræmia, due to nephrocalcinosis, due to parathyroid tumour.

Figs. 251-253 show the presence of abnormal calcification in the lungs, a vessel in the kidney, and in the kidney substance itself, respectively.

Only 12 cases of carcinoma of the parathyroid with metastases have been reported in the literature; a further 10 or 11 cases reported as carcinoma have not been fully verified, because the criteria for the diagnosis of carcinoma of this gland have not been fully established. It is felt by some that an unequivocal diagnosis of carcinoma can be made only if metastases are found, either at the time of examination or at a later date. It is believed, however, that a tumour which is densely adherent or locally invasive at the time of the original operation, and subsequently recurs, is also carcinoma. Parathyroid carcinomata are usually larger in size than the adenomata, and for this reason are more frequently palpable than the latter, prior to operation. The microscopical appearance can at times be quite confusing, because an adenoma may show extremely pleomorphic cells and nuclei, giant cells, nests of cells, within the surrounding capsule, and small nests of tumour cells lying free within blood-vessels. Castleman (1952) feels, however, that all tumours of the parathyroid that show mitotic figures are carcinoma, no matter how infrequent the mitoses may be. He also believes that a trabecular arrangement of the tumour cells is significant in the diagnosis.

The present case conforms with the histological criteria of carcinoma of the parathyroid and the post-mortem findings suggest the possibility of local recurrence after excision. In the absence of absolute proof as provided by the presence of metastatic lesions it must be admitted that the case is "not proven".

DISCUSSION

The clinical features of the fully established case of hyperparathyroidism with bone changes, be it due to hyperplasia or tumour formation, are nowadays too well known to merit further repetition. There are certain points, however, particularly in the early diagnosis and in the prognosis of the disease, that are perhaps not so familiar, which we might be permitted to stress.

Commonly in the early stages of the disease there are vague complaints the nature of which is unlikely to be recognized unless the possibility is in mind or unless it is suggested by some fortuitous happening or investigation. The early symptoms consist principally of ill-defined pain in the abdomen and limbs, anorexia, weakness, lassitude, polydypsia, and polyuria; vomiting may occur. These symptoms are believed to be caused by a raised serum-calcium level. Peptic ulceration and nephritis are frequently suspected and, indeed, may be present and demonstrated by suitable investigation. Once such a diagnosis is justifiably made, the case is likely to be treated along the standard lines prescribed for these conditions and further investigation abandoned. On the other hand no definite organic lesion may be found and the patient may be labelled 'neurotic' and sedatives will be ordered until the progress of the disease confronts the attendant with an obviously ill patient. It is possible that during the performance of radiological studies of the alimentary canal or of the renal tract abnormalities may be detected in the neighbouring skeleton, thus giving an unexpected clue to the true nature of the disease present. Suspicion may also be aroused by the presence of bilateral renal calculi, or by the presence of widespread soft-tissue calcification. The expectations of such fortuitous happenings must not, however, be relied upon, for symptoms of abdominal pain and polyuria may precede by many years the onset of bony abnormalities. Furthermore, it is held by some authorities that in 35 per cent of cases of hyperparathyroidism no bone changes are present. It is therefore well worth while to remember the possible presence of hyperparathyroidism in chronic cases of ill health which are characterized by anorexia, lassitude, abdominal pain, polydypsia, and polyuria. An unexplained nephritis may also be profitably investigated in this direction. This is well exemplified by *Cases* 2 and 3.

The importance of an early diagnosis cannot be sufficiently stressed because of its important bearing on prognosis. In the advanced stage of the disease, in spite of the correct diagnosis having been made and adequate treatment carried out, the extent of the nephritis usually present is commonly too advanced to be substantially altered. Death from renal failure within a relatively short time of operation is, unfortunately, a common enough occurrence. This was the case in our third patient, which should be contrasted with the first. Furthermore in those instances where bone changes are present, although the abnormal osseous metabolism may be halted and recalcification slowly obtained in the wake of effective treatment, the deformities present may well be too extensive to be corrected and will remain as a permanent legacy.

The importance of recognizing the coexistence of peptic ulceration in cases of hyperparathyroidism must be emphasized anew. In such instances treatment with the standard diets and antacids will lead to a heightened calcium intake which might well push the patient into an acute stage of hyperparathyroidism with possible fatal results (Robinson, Black, Sprague, and Tillish, 1951; Rogers, 1946; Rogers, Keating, Morlock, and Barker, 1947). The problem is a difficult one, for the abdominal pain, nausea, and vomiting sometimes associated with hyperparathyroidism *per se* may closely mimic the symptoms of peptic ulceration. The onset of persistent vomiting and deterioration in peptic ulcer cases undergoing adequate treatment, and in which no obstruction in the

alimentary tract can be detected, should make one suspect such an unhappy association. Alkali and milk therapy should be stopped at once and suitable investigations made.

It will have been noticed that in spite of the extensive bony changes present in the 3 cases that we have reported, in each instance the level of the alkaline phosphatase in the serum was normal or occasionally raised to a very slight degree. This is a not infrequent observation in osteitis fibrosa cystica of the generalized type (Robinson and others, 1951). It should also be stressed that a single estimation of the serum-calcium may be misleading. The serum-calcium levels can fluctuate widely in hyperparathyroidism and repeated assays are advisable. It is a curious fact that the level of hypercalcæmia varies very markedly from one case of hyperparathyroidism to another with an apparently similar lesion; thus at times the serum level may be as high as 20 mg. per cent and at others but a little above the upper level of normality. The serum-phosphorus, however, is constantly depressed.

Some authors claim that in the presence of a negative Sulkowitch test the possibility of hypercalcæmia need not be considered any longer. This may well be true, but in our first case only a trace of calcium was repeatedly present in the urine. Such a trace may be present in the normal healthy person. Again, repeated estimations, preferably of twenty-four hour specimens are advisable.

As regards the operation of parathyroidectomy we would stress certain desiderata. A wide comfortable incision is an essential. Absolute hæmostasis must be ensured throughout; vessels must be scrupulously ligated and divided as they are encountered. This applies particularly to cases where previous neck surgery, e.g., thyroidectomy, has been performed. Careful search must be made of all the known locations of the parathyroid glands; until then, no tissue should be excised. It must be remembered that the presence of one adenoma does not exclude the presence of others; more than one tumour is said to occur in 10 per cent of instances. *Case* 2 is one such example. In the presence of hyperplasia it is wise to know how much parathyroid tissue is present before embarking upon a wholesale ablation if troublesome hypoparathyroidism is to be avoided. Worst of all, if success is claimed prematurely, normal parathyroid tissue may be removed and an adenoma left behind. The size of an adenoma may be extremely variable; it may be obvious, or it may be so small as to be hardly countenanced as a tumour. Thus in our first case the left superior parathyroid gland was removed, not because we were convinced that it was a tumour, but rather because it represented the only parathyroid tissue recognizable. Most parathyroid tumours will be resectable by the standard approach used in thyroidectomy. Sternotomy is not often required; thyroidectomy has been recommended where all other lines of attack have failed to detect the culprit, for the parathyroid glands may at times be embedded in the thyroid gland itself.

SUMMARY

Three unusual cases of hyperparathyroidism have been presented. One occurred in a 3-year-old child; the second was characterized by the peculiar association of recurrent hyperparathyroidism with recurrent peptic ulceration; whilst in the third the histological findings are highly suggestive of malignancy. Some of the less well-known features of hyperparathyroidism have been reviewed.

Acknowledgements.—It is with great pleasure that we acknowledge our indebtedness to Dr. H. H. Pitts, Pathologist at St. Paul's Hospital, Vancouver, for his inestimable help and advice. To our many colleagues at the C. S. Williams Clinic and to our more recent colleague, Dr. H. Duggan, Pathologist at the Trail-Tadanac Hospital, we wish to express our gratitude for the many and varied contributions that they have made to our work.

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'POLYPOSIS' OF THE GALL-BLADDER

By D. J. OAKLAND, Birmingham

Although the medical literature contains many papers describing 'papillomata' of the gall-bladder, it is doubtful whether some of these lesions are truly neoplastic in nature, benign tumours of the gall-bladder being regarded by most authorities as rare lesions (Kerr and Lendrum, 1936; Brown and Cappell, 1937; and Willis, 1948). The 2 cases of polyposis of the gall-bladder which are described below, although similar radiologically and macroscopically, represent two types of polyp which are found in the gall-bladder.

Fig. 254.—*Case* 1. Cholecystogram.

Fig. 255.—*Case* 2. Cholecystogram.

CASE REPORTS

Histories.—

Case 1.—H. T., a female patient of 61, consulted her doctor on account of an increase in her weight. Routine examination revealed marked localized tenderness in the right hypochondrium, and on further questioning she admitted to occasional attacks of nausea and vomiting. A cholecystogram showed several negative shadows in a functioning gall-bladder (*Fig.* 254). These shadows appeared to arise from the wall of the gall-bladder inasmuch as they were not surrounded on all sides by the radio-opaque material. Laparotomy revealed a gall-bladder of normal appearance, but small nodules could be palpated within its lumen. The other abdominal and pelvic viscera appeared normal. Cholecystectomy was performed, and examination of the gall-bladder revealed many small excrescences arising from the entire mucous surface of the organ. These proved to be polypi and varied between 4–10 mm. in height. There were no calculi and the mucous membrane did not appear diseased on macroscopical examination.

Case 2.—D. M., a young general practitioner, who was admitted as an emergency with a ten-day history of upper abdominal pain coming on after meals. The interval between food and pain had decreased until two days prior to admission, when the pain had come on during a meal and had been very severe throughout that day. The patient had been able to carry on with his work. On examination there was tenderness and guarding in the right hypochondrium, and the patient had a temperature of 99·2°. A diagnosis of resolving acute cholecystitis was made and the patient was kept under observation. Cholecystography performed seven days after admission demonstrated a functioning gall-bladder which contained several negative shadows in its lumen (*Fig.* 255). These shadows appeared to arise from the wall of the gall-bladder and were diagnosed as polypi. Laparotomy was performed and a chronic duodenal ulcer was found, which appeared to have recently leaked and had become sealed off by omentum. The gall-bladder appeared normal, but palpation revealed several nodules within its lumen and cholecystectomy was performed. Examination of the organ revealed numerous small polypi without other evidence of disease.

Histopathology.—

Case 1.—The mucosa of the gall-bladder showed diffuse papillomatous hyperplasia. The papillomata were covered with columnar epithelium, deep to which was a fibrovascular stroma heavily infiltrated with lipid-containing histiocytes ('foam cells'), the

cytoplasm of which appeared clear in paraffin sections (*Figs.* 256, 257). Doubly refractile material was demonstrable on polarization microscopy. The submucosa showed mild infiltration with chronic inflammatory cells.

Case 2.—Apart from a number of small papillomata the mucosa of the gall-bladder was comparatively normal in appearance. Each papilloma consisted of

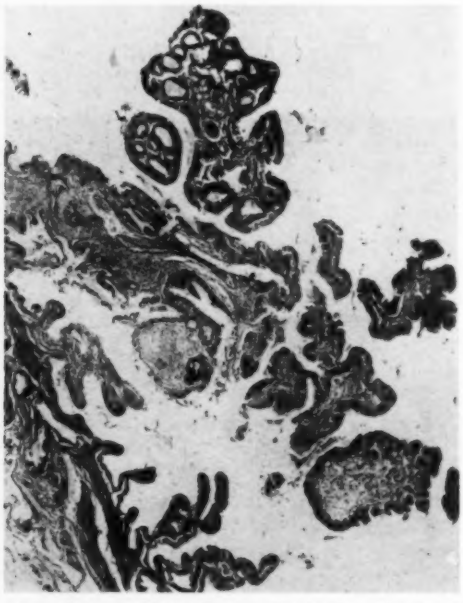

FIG. 256.—*Case* 1. Photomicrograph of polyp of gall-bladder showing infiltration with cholesterol. (× 28.)

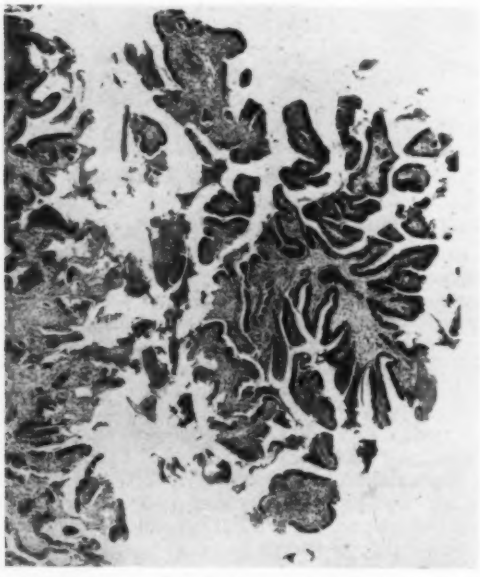

FIG. 258.—*Case* 2. Photomicrograph of papilloma of gall-bladder. (× 28.)

a complex mass of fronds lined by columnar epithelium, which covered a sparse fibrovascular stroma. There was slight chronic inflammatory-cell infiltration in the stroma of the papillomata and in the uninvolved gall-bladder wall (*Fig.* 258).

DISCUSSION

The medical literature contains many excellent descriptions of papillomata of the gall-bladder. However, some confusion has resulted from the fact that many of these papillomata are not true neoplasms, as the name implies. The term 'papilloma' has been applied to polypoid conditions of the gall-bladder which are histologically similar to the

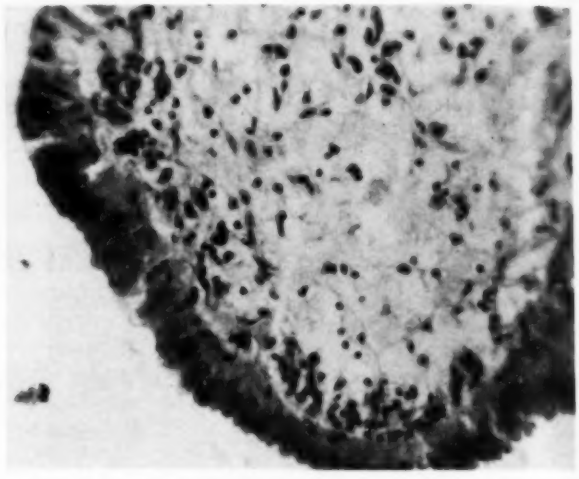

FIG. 257.—*Case* 1. Photomicrograph showing lipid-containing histiocytes—'foam cells'. (× 198.)

cholesterol laden villi of the strawberry gall-bladder. For this reason figures on the incidence of papilloma of the gall-bladder vary considerably. Walters and Snell (1940) state that innocent papillomata occur in 8 per cent of surgically removed gall-bladders. Kerr and Lendrum (1936) made a critical analysis of cases reported in the literature as papilloma of the gall-bladder up to 1936 and found that they could accept only 7 cases as true papilloma. Kane, Brown, and Hoerr (1952), in a review of the records of the Cleveland Clinic since 1921, found only 8 papillomas in 2000 gall-bladders which had been removed during this period. They pointed out that during the same period there had been 30 cases of carcinoma which is itself regarded as an uncommon disease. Kaufmann (1922) believes that the gall-bladder epithelium becomes more papillomatous with increase of age. Phillips (1933) examined 500 cases of papillomata of the gall-bladder which had been found in gall-bladders removed at the Mayo Clinic between 1923 and 1929. He distinguished two varieties :—

1. The inflammatory type which had many projections and little stroma and was found in thick-walled badly infected gall-bladders.

2. The inflammatory and metabolic type which were localized collections of lipids, including cholesterol, in the submucosa and deeper parts of the epithelium of the gall-bladder to such an extent that a polypoid excrescence was formed. He pointed out that many of the cases described in the literature belonged to this latter group, which was one stage in the well-known condition of strawberry gall-bladder. In this classification he did not include a true papilloma comparable with the papilloma found in the urinary bladder or the alimentary tract.

As Tabah and McNeer (1953) point out in a recent paper, neither of the types of papilloma described by Phillips is strictly neoplastic. The inflammatory group occurs in badly infected gall-bladders where there has been active proliferation of the epithelium of the mucosa with the formation of polypoid projections. In some cases the proliferation develops downwards, with the result that the mucosal projection penetrates the muscularis, forming a Rokitansky-Aschoff sinus. King and MacCallum (1931) have suggested that this type of inflammatory polypoid disease of the gall-bladder would best be described by the term ' cholecystitis glandularis proliferans '.

Some authors (Shepard, Walters, and Dockerty, 1942) maintain that true papillomata do not occur in entirely normal gall-bladders, and found some evidence of chronic inflammation in every one of their cases from the Mayo Clinic.

The first of the cases described above corresponds to the inflammatory and metabolic type of Phillips. The generalized polypoidal state of the mucous membrane, heavily infiltrated with cholesterol, is similar to the appearance of a strawberry gall-bladder, the polypi being merely an exaggeration of this process in places. Further deposition of cholesterol in these papillomata would eventually lead to a similar macroscopical appearance to that of a typical case of cholesterosis of the gall-bladder shown in *Fig.* 259. In this case masses of cholesterol were attached to the wall of the gall-bladder by fine pedicles.

The second case is a true papilloma and fulfils the requirements of Tabah and McNeer (1953). However, there is some round-cell infiltration of both the papilloma and the gall-bladder wall, which suggests an inflammatory factor in the aetiology of this lesion.

Many authors agree (Phillips, 1933, and Kane and others, 1952) that true papillomata of the gall-bladder are unlikely to give rise to symptoms, although Tabah and McNeer (1953) state that the patient's symptoms are very similar to those of chronic cholecystitis with or without stones. In the second case described the papilloma was discovered accidentally and this patient's symptoms were undoubtedly due to his leaking duodenal ulcer. In the first case the papillomata were part of a generalized cholesterosis of the gall-bladder, and the patient's symptoms and signs appeared to be more definitely related to the state of this organ.

In both these cases the diagnosis was suggested by the cholecystographic appearance before operation, but usually operation is performed with a diagnosis of cholelithiasis. Kirklin (1931, 1933), who was the first to demonstrate papillomata by cholecystography, claims that the diagnosis can be made with considerable accuracy by this technique. His view is supported by Moore (1935), who stresses the importance of oblique views.

The significance of papillomata of the gall-bladder is important. If the diagnosis is made radiologically during the course of investigation of a patient's symptoms, what action should the surgeon take? The relationship between papillomata and carcinoma has been extensively discussed by Phillips (1933), and in his series there was only one papillomatous gall-bladder, which contained a carcinoma, and in his case there was no evidence that the carcinoma had arisen in one of the papillomata. In his view the presence of papillomata in a gall-bladder signified a lesser degree of inflammation and correspondingly less injury to the gall-bladder mucous membrane. On the other hand, carcinoma of the gall-bladder commonly arises in an organ which has been the seat of severe chronic inflammation for a long time, and calculi are almost always present.

Shepard and others (1942) also state that they could find no well-authenticated instance of malignant change in papilloma of the gall-bladder in the medical literature. However, Tabah and McNeer (1953) report 4 cases of true papillomata of the gall-bladder, with histological evidence of carcinomatous change in 3 of them. They claim they are the first recorded and proved examples of malignant change occurring in a papilloma of the gall-bladder.

FIG. 259.—Photograph of gall-bladder polyps containing cholesterol.

There seems to be a more definite relationship between papillomatosis of the gall-bladder and gallstones. This relationship is particularly obvious in those cases where the papillomata are partly metabolic in type, but even if a purely inflammatory or true papilloma of the gall-bladder is broken off, it will form an excellent nidus for the formation of a stone (Boyd, 1948).

Most authorities agree that although there is no definite evidence that true papilloma of the gall-bladder is the precursor of carcinoma, it is reasonable to assume that if benign papillomata in other organs can undergo malignant change, then this may occur in the gall-bladder. The recent reported cases of Tabah and McNeer (1953) would seem to support this view. Therefore, it would appear that cholecystectomy was indicated once the diagnosis of papillomatosis of the gall-bladder had been established.

SUMMARY

Two cases of papillomatosis of the gall-bladder have been described. The pathology of this disease and the incidence of true papilloma has been discussed. The relationship of papilloma to carcinoma and calculous disease of the gall-bladder has been considered.

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THE BILLROTH I GASTRECTOMY

BY PAUL HICKINBOTHAM, LEICESTER

RENEWED interest has been taken in the Billroth I type of gastrectomy in recent years. This operation, which involves restoration of continuity after the gastric resection by gastroduodenal anastomosis, was abandoned in the early days of gastrectomy because of a tendency to leakage at the suture line ; this danger has now largely been overcome, probably because of a greater appreciation of the two fundamental principles : (1) only to suture tissues which have a good blood-supply ; and (2) never to suture under tension. Many favourable reports of this operation have now been published ; Schmitz, Harkins, Moore, and Olson (1954), for instance, are enthusiastic protagonists of it and suggest that the operation, as done by them, is applicable to most cases of duodenal ulcer ; only in the few cases in which the operation was technically not feasible did they apparently resort to a Polya type. They reported that the result was satisfactory in 92 per cent of cases.

As we at the Leicester Royal Infirmary had not had the same experience, it was thought that it would be worth while to review all the Billroth I gastrectomies carried out in the hospital up to 1952, thus allowing a minimum follow-up period of two years. The series is complete, though it covers a period during which the operation was gaining popularity and there were, therefore, many Polya operations being done for the more difficult duodenal ulcers. The cases were operated on by six surgeons.

The series comprises 276 cases, of which 6 were cases in which the gastrectomy was the primary treatment of a perforated peptic ulcer and 15 cases in which the operation was one of urgency for an actively bleeding ulcer. The results have been divided into three categories : (1) satisfactory, having no digestive symptoms or only such as might be expected in a normal person ; (2) improved, in which the patient has definitely derived benefit from the operation, is doing a full time job, but does have some symptoms referable to his digestion ; (3) failures—this group includes known or suspected recurrent ulcers, anastomotic stenosis, and patients who for other reasons feel that they have not benefited from the operation. Any classification is subjective and of necessity inaccurate ; this paper, however, is mainly concerned with the recognizable conditions of recurrent ulcer, stenosis at the suture line, and the ' dumping syndrome '.

RESULTS

(*Table I*)

The cancer cases present a gloomy picture and in many of them the operation was only a palliative procedure. There were 3 operative deaths : a lady of 75 who developed the clinical picture of bronchopneumonia and died 5 days after operation, no autopsy being done ; a man of 58, who had a palliative resection and who died 5 days later of what seemed clinically to be a leaking suture line—his histological section showed that the growth was more extensive than was thought at operation, being in fact a linitis plastica extending right up to the line of section of the stomach ; the third case was also a palliative operation and death occurred 4 days later—at autopsy leakage from avascular necrosis of a small area of the greater curve of the stomach at the anastomosis was found. Of the 14 who have since died, recurrent cancer has been the cause in 12, the average survival being 11 months. Three remain alive and free from evidence of malignant disease 44, 41, and 36 months after operation ; 1 is alive, but with recurrence, at 50 months. The 1 untraceable patient was known to be alive at 43 months because she attended the Dermatological Department with a contact dermatitis of her neck from wearing a fur collar.

The majority of cases were operated on for benign peptic ulcers, and in these there were 7 operative deaths ; in each case the diagnosis was confirmed by further operation or by autopsy. They were as follows : 1 gastric ulcer case—leakage at the lesser curve suture line ; 5 duodenal ulcer cases—avascular necrosis of an area of duodenum, separation of the suture line, lung abscess resulting in death 2½ months after gastrectomy, coronary occlusion, and 1 case of uræmia. This last man had a blood-urea of 70 mg. per cent before operation, and this progressed until it reached 376 mg. per cent when he died three weeks later. The gastric and duodenal case died from coronary occlusion.

The essential facts elicited by the follow-up are shown in the table. Throughout the series it was found that a sensation of fullness after meals was a common, almost universal, occurrence in the early post-operative months ; it usually improved with time, but this was not invariable ; when persistent, however, it was never severe and seemed to

cause little trouble. The other common finding was that though most of the patients had regained their pre-operative weight, 32 per cent had not regained their normal weight prior to the development of ulcer symptoms. This, while being of interest, is probably not important and no case of serious malnutrition was seen. These two symptoms—postprandial fullness and failure to regain weight—are presumably the mechanical result of a small stomach.

There were 7 cases of definite early postprandial syndrome: patients who felt tired, sleepy, and useless after a meal—usually the main meal of the day—and who sometimes had to lie down and go to sleep; some complained of sweating as well. These symptoms seemed more pronounced if fluid was taken with the meal and some had learnt to take their fluids at other times. One case had a late postprandial syndrome; he felt faint and weak and sweated if he went too long without food, the symptoms being readily cured by a meal.

In any large series of peptic ulcer patients there are bound to be failures owing to the patient's poor psychological background; such a case is one lady on whose notes the physician wrote before the operation "If this lady had all the postgastrectomy syndromes she could not be worse than she is at present." Such cases will have a high failure rate, but it does not seem right to refuse operation to a patient with a peptic ulcer simply because he or she has a neurotic personality. Other patients who appear in the failure group are those whose symptoms may in fact not have arisen from the peptic ulcer in the first place; one such man was noticed at the operation to have a nodular pancreas and as his symptoms have remained very similar the question of chronic pancreatic pain arises.

The general impression gained in interviewing these patients, however, was that the great majority were either very well and extremely grateful or had some organic complication such as recurrence or stenosis. There were few of the rather puzzling, not entirely satisfactory cases which one seems to see more commonly after the Polya operation: the follow-up was, in that sense, easier—either they were well or there was something definite the matter.

Recurrences.—One of the 132 gastric cases is recorded as a recurrence although the symptoms are not quite typical; a barium meal shows what is probably a recurrent ulcer on the duodenal side of the anastomosis and he is awaiting laparotomy. Of the 100 cases of duodenal ulcer, however, there are 10 recurrences; of these, 7 have been confirmed by operation and vagotomy performed with very satisfactory results; 1 case has been confirmed and converted to a Polya; 2 cases are suspected, but the symptoms do not at present justify a further operation. Symptoms of recurrence started at times varying from 3 months to 4 years after gastrectomy, the average being 2 years. The significance of this is clear: our follow-up of the majority of the cases was less than 4 years, though in all it was over 2 years. This must mean that we may expect further cases now classified as satisfactory to move into the failure group with recurrences. The final figure will therefore be well above 10 per cent.

Table I.—Analysis of Cases treated by Billroth I Gastrectomy

Type of Case	Number	Operative Deaths	Since Died	Untraced	Success	Improved	Failure	
							Recurrences	Others
Gastric	132	1 (0·8 per cent)	12	7	97 (86·7 per cent)	14 (12·5 per cent)	1 (0·8 per cent)	—
Duodenal	100	5 (5·0 per cent)	4	4	64 (73·6 per cent)	6 (6·9 per cent)	10 (11·5 per cent)	7 (8·0 per cent)
Duodenal and Gastric	19	1 (5·3 per cent)	2	—	15 (93·7 per cent)	—	—	1 (6·3 per cent)
Cancer	22	3 (13·6 per cent)	14	1	3	—	—	1
No ulcer	3	—	—	—	—	2	—	1

In passing, it may be pointed out that a recurrent ulcer is almost always a duodenal ulcer and has not the dangerous potentialities of a gastrojejunal ulcer. One is dealing, in fact, with a problem similar to that with which the patient presented with his original primary ulcer.

Stenosis.—There were no cases of stenosis in the gastric ulcer group; there were, however, 2 cases in the duodenal group, both of which have required operative treatment. This relatively low incidence is gratifying as it was one of the complications which some of us feared might arise more frequently.

DISCUSSION

The Billroth I operation has many advantages over the Polya: it restores the gastro-intestinal tract to a more nearly normal condition both anatomically and physiologically. The various mechanical hazards associated with a loop—afferent loop obstruction, burst duodenal stump, volvulus, intussusception, etc.—are eliminated. The bile and pancreatic secretions enter the digestive tract in the normal sequence and mix fully with the food. The possible disturbances caused by a by-passed section of bowel are avoided. As a result, the functional results of the operation are, I think, considerably better than those of the Polya; this is a view which is becoming increasingly widely held by surgeons with experience of both types of gastrectomy. Capper and Welbourn (1954) say "It is clear that the Billroth I procedure gives a much better functional result in nearly all respects than do operations with gastrojejunal anastomoses".

How often is a Billroth I gastrectomy feasible and how safe an operation is it? The cases here reported give no idea of how often the operation was done in preference to the Polya type: my personal cases done during the two years since the present review are as follows: 80 duodenal ulcers, 47 gastric ulcers, 8 combined duodenal and gastric ulcers, and 10 cancers. Of these, out of the total of 145 cases,

all but 2 were Billroth I operations; these latter 2 cases were duodenal ulcers and Polya type gastrectomies were performed. This shows that there are very few cases in which with care and persistence a Billroth I operation is not possible.

The operative mortality is shown in *Table I*. It will be seen that for gastric ulcers it was under 1 per cent, whereas for duodenal ulcers it was 5 per cent. This latter figure is high, but it must be remembered that the series included the early Billroth I operations done in the hospital; a number of technical points were not fully appreciated at that time and the mortality now is considerably less. In my own subsequent 145 gastrectomies there have been 2 deaths; this series excludes 15 additional cases done for hæmatemeses, in which there have been 3 deaths. It would seem therefore that the Billroth I operation is at least as safe as the Polya, and I personally think that it is safer.

As regards technical points learnt, the two vital factors are that there must be no tension and that the stomach and duodenum must have an adequate blood-supply. The former can be ensured by mobilizing the greater curve of the stomach fully, so that if necessary only the last vas brevis is left. The blood-supply of the stomach is usually excellent but the duodenum requires great care; I believe that it must not be dissected off the pancreas to give a comfortable length for suturing; this is to invite avascular necrosis. It is better to do a difficult and less neat anastomosis to good vascular duodenum. Given no tension and vascular parts, leakage from any suture line is most unlikely unless there is pressure within the organ: we are all aware, for instance, that closure of a perforated duodenal ulcer is almost always watertight, although the suturing may be rather crude; whereas the best closed duodenal stump will leak if there is afferent-loop obstruction. Thus, with attention to the points mentioned, leakage is not to be feared with Billroth I operation.

This review shows clearly that the operation has been overwhelmingly successful in the treatment of gastric ulcers and I believe that at the present time it is the operation of choice. If it be true, as suggested by Dragstedt, Oberhelman, Evans, and Rigler (1954), that the important factor in the aetiology of gastric ulcers is an oversecretion of gastrin by the pyloric mucous membrane, then a low Billroth I gastrectomy is the rational treatment.

Our duodenal ulcer cases, however, showed an incidence of recurrent ulceration which is disturbingly high. This is not due, as is often suggested, to an inadequate resection occasioned by the necessity of joining the stomach back to the duodenum. With adequate mobilization of the stomach, the resection can be as high as is required; this has been confirmed in several cases in which a Polya has subsequently been converted without difficulty into a Billroth I gastrectomy. The real reason for the increased frequency of recurrences is, I believe, this: a simple gastrojejunostomy will cure the majority of duodenal ulcers; if to this is added a gastric resection the cure becomes more certain and a Polya operation has been performed. It is this combination of gastrojejunostomy effect with gastric resection which makes the Polya operation proof against recurrences; the Billroth operation, lacking the gastrojejunostomy component, is prone to recurrences.

It is my view that the Billroth I operation is not suited to the treatment of duodenal ulcers; the treatment which I am now adopting is a vagotomy combined with a low Billroth I gastrectomy. This seems better than a vagotomy combined with a gastrojejunostomy or pyloroplasty because of the relatively common coexistence of a gastric with a duodenal ulcer; under these circumstances one might very well miss a small gastric ulcer and it might cause subsequent trouble unless the pyloric half of the stomach were removed. Such an experience has now occurred to me on more than one occasion—the specimen after gastrectomy for a duodenal ulcer revealing an unexpected gastric ulcer.

My thanks are due to my colleagues at the Leicester Royal Infirmary for permission to use their cases, though the opinions expressed in this paper are purely personal.

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EXPERIMENTAL SURGERY

STEREOSCOPIC MICRO-ANATOMY

By A. G. PARKS, London

Shortly after the invention of the microscope, a great interest in the minute structure of tissues developed, and because there were no microtomes, freehand sections were cut of about 100μ or more thick. It is surprising to find just how much was discovered with relatively crude techniques. Some early nineteenth century text-books of histology are full of interesting and accurate descriptions of minute glandular structure and vascular patterns. Microtomes were used towards the end of the century which enabled sections to be cut as thin as 5μ. As a result cellular structure became more easily visible and the emphasis of study so shifted that cytological histology almost completely ousted the science of tissue architecture. The appeal of very thin sections has so blinded us that even to-day we try to conjure up a three-dimensional mental picture of an organ by studying a few sections which have two dimensions only. Needless to say we usually fail, but our failure goes uncorrected.

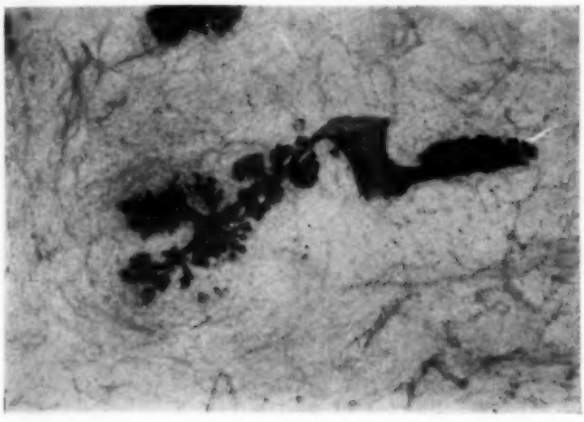

Fig. 260.—Major breast duct with terminal branches. (Thick section.) (× 10.)

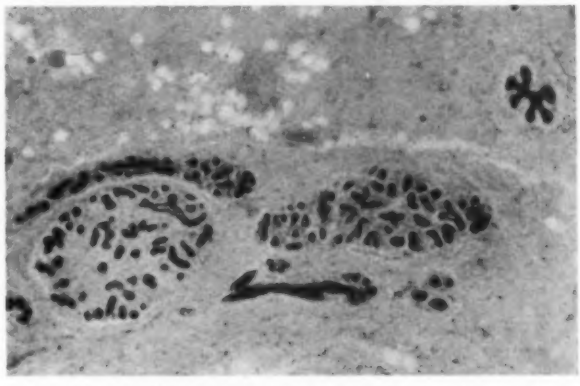

Fig. 261.—Section similar to *Fig.* 260, but cut as thin section. (× 15.)

Our knowledge of gross anatomy and of cytology has progressed steadily. There is still a realm between the gross and the minute which is relatively unexplored; it consists of the micro-anatomical structure of organs, especially the pattern of glandular tissues and minute blood-vessels. So that a clear concept of an organ such as a gland can be obtained, it is necessary that a sample of its ducts and acini be seen in continuity. It is possible to build up models from thin sections, but the work is very time-consuming and tedious. The only practical solution is some method whereby a thick slice of tissue may be examined.

In the past certain very thin structures have been mounted whole, for instance the breast of the mouse (Jull, 1954), the choroid plexus (Millen and Woollam, 1954) and organs of invertebrates. Thick sections cut on a freezing microtome have been extensively used especially in the examination of the central nervous system by silver techniques. There are limitations to both these methods.

There are two technical difficulties involved in cutting thick sections. The first is to find an embedding medium which will support the tissue, yet will not be too hard for the knife to cut. It is difficult to cut a thick paraffin section and the wax is dissolved in the process of mounting so that the section cannot be labelled by imprinting a number on it. Celloidin, which is nitrocellulose of high viscosity, penetrates tissues poorly and does not form sufficiently concentrated solutions. Chesterman and Leach (1949) found low viscosity nitrocellulose (the substance with which cars are sprayed) to be much more satisfactory than either of the preceding. It penetrates fairly rapidly; a solution containing 20 per cent nitrocellulose can be obtained easily and the final block is hard enough to allow thick sections to be cut from it.

The second problem was to find a stain which supplied contrast to the various elements of a tissue, yet rendered it translucent enough to allow vision through a certain thickness of the section. Ordinary stains such as hæmatoxylin and most of the aniline derivatives turn a thick section into an opaque mass. Leach (1952) found anthracene blue to be the only satisfactory dye. It is a nuclear stain which has a strong affinity for epithelial structures. Nerves and smooth muscle, including blood-vessels, are faintly stained; fat, fibrous tissue, and striated muscle are hardly stained at all. For best results the final section should consist of faintly stained material through which the more deeply staining epithelial structures pass. The breast is the ideal organ to study with this method as the brightly stained ducts run through colourless fat and fibrous tissue. Organs composed almost entirely of epithelium, for instance the liver and kidney, are unsuitable as they are opaque and nearly black after staining. However, the minute vascular pattern of any organ can be

satisfactorily studied with these thick sections if the vessels are first injected with indian ink or some other injection mass. I have found the method of Millen and Woollam (1953) to be excellent for this purpose. Two coloured suspensions of different particle size are injected into the arterial tree. The finer suspension passes through the capillaries into the veins; this is followed by the more granular injection which

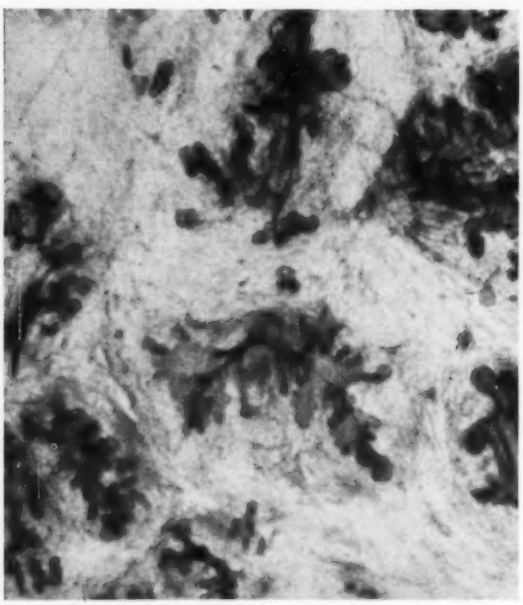

FIG. 262.—Breast duct showing laminar growth. (Thick section.) (× 15.)

will not enter the capillaries. It is usually unnecessary to stain the tissue as well but it may help to stain it very lightly so that the vessels are seen coursing around the ghost-like glandular structures. In order to obtain a three-dimensional effect in the section it is necessary to use a stereoscopic binocular microscope of the Greenough type. Magnifications from 5 to 50 can be used; above this the depth of focus becomes so small that three-dimensional vision is lost.

Perhaps the greatest potentialities of this method are to be found in teaching. It may be good intellectual training for a student to have to build up structural concepts of organs by fusing together many thin slices, but it would surely be far better if structure were perceived directly. With thick sections the student can see a sweat-gland in its entirety or the greater part of a breast duct with its terminal branches. He does not have to struggle to build a three-dimensional concept from two-dimentional slides, a task which is almost impossible for a complicated structure such as, for instance, an intracanalicular fibro-adenoma.

Thick sections can be used for research purposes and are especially valuable in the complete histological examination of a block of tissue, a task usually performed by the use of serial paraffin sections. A piece of tissue 5 cm. thick can be completely surveyed by one hundred thick sections, whereas 1000 thin paraffin sections would be required. A thick section can be stamped with a number in indian ink and this is indelibly printed on it, even when mounted, as the nitrocellulose is deliberately preserved and is not dissolved away from the section. There are thus none of the difficulties met with in attempting to mount long ribbons of thin sections. Indeed, thick sections do not have to be mounted at all unless it is especially desired. They can be examined in a dish of mounting medium and in this way all the work which normally goes into mounting is avoided. They are conveniently stored in a very compact fashion in small jars. Even if it is required to mount them, it is a much easier task than mounting large thin sections cut from celloidin or paraffin wax. Finally, if any area in the thick section is suspect or

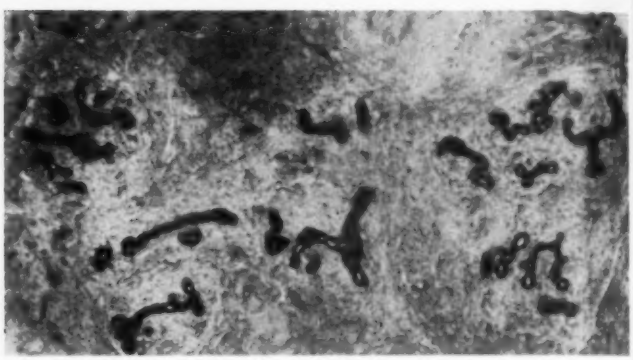

FIG. 263.—Thin section of tissue adjacent to the area photographed in *Fig.* 262. This section suggests that ducts are being cut longitudinally. In fact these are laminar ducts cut transversely. (× 15.)

of especial interest, it can be punched out with a paper punch, embedded in paraffin wax, and cut in as thin a section as is required. In this way the advantages of two techniques are combined. A large amount of tissue can be scanned in a short time. Once found, the object of the search may be examined cytologically.

By this method a whole realm of minute anatomy is revealed and many applications suggest themselves. The study of breast structure has been most helpful to the understanding of the pathology of fibro-adenoma and fibro-adenosis. A similar investigation of the terminal bronchioles, the salivary glands or the prostate might well be equally revealing. Neoplastic change can be identified with certainty provided that it is of the infiltrating variety. It is recognized by the way in which it destroys the orderly arrangement of ducts and glands, and by the tongues of malignant tissue radiating from the body of the tumour. In this method there are no difficulties due to tangential section of ducts as occurs occasionally with thin paraffin sections. If the tumour does not infiltrate it is seen as a darkly stained mass of epithelial tissue which distorts, but does not destroy normal anatomy (for instance, intraduct carcinoma of the breast). This variety is indistinguishable in a thick section from benign epithelial hyperplasia. In order to differentiate between these kinds of epithelial proliferation, a segment is punched out of the thick section and a paraffin section is prepared from it. Though carcinoma is too densely epithelial to be studied with much profit, growth in lymphatics or in ducts is readily seen and may be traced throughout its course. When combined with an injection technique thick sections enable the minute vascular

pattern of an organ to be seen clearly. These nitrocellulose sections are easier to manipulate than frozen sections which are usually used to examine injected material.

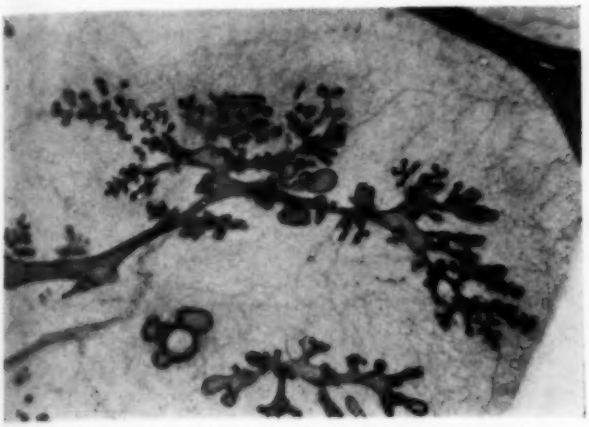

FIG. 264.—Thick section of breast tissue showing early dilatation of terminal ducts. (× 10.)

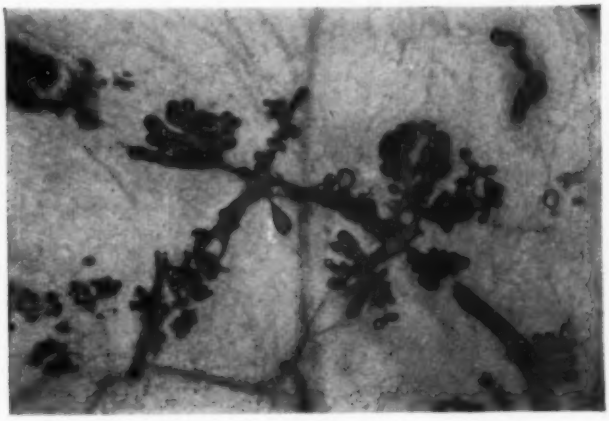

FIG. 265.—Thick section showing similar cystic changes. (× 10.)

The accompanying photographs show some of the results obtained, though a black and white print gives a poor representation of the original which is seen with three-dimensional perception.

TECHNIQUE

Modified from Chesterman and Leach (1949) and Leach (1952).

Fixation.—Specimens are fixed in 10 per cent formol saline for one or more days according to size.

The following is a scheme which has been found to be generally useful :—

Alcohol per cent	*Size of Specimen*		
	1–2 *cm.*	2–5 *cm.*	5–20 *cm.*
30	1 day	2 days	3 days
70	1 day	2 days	3 days
90	1 day	2 days	3 days
Abs 1	1 day	2 days	1 week
Abs 2	1 day	2 days	1 week
Abs 3	1 day	1 week	1 week

Dehydration must be complete before low viscosity nitrocellulose will penetrate.

Defatting.—Very fatty specimens should be treated with benzol or xylol until clear of fat.

Impregnation.—Low viscosity nitrocellulose (L.V.N.) is obtained in the form of a light, fluffy powder ; it is damped with butyl alcohol to decrease its great inflammability. Like celloidin it is soluble in a mixture of alcohol and ether, is softened and distorted by absolute alcohol, but is not affected by isopropyl alcohol. It forms a tough block which can be cut in the dry state. A small proportion of ordinary celloidin is added to the L.V.N. to reduce brittleness of the final block. L.V.N. impregnates fairly rapidly from 5 per cent and 10 per cent solutions made up in a mixture of equal parts of ether and absolute alcohol. Alcohol by itself wets the material and, on stirring, a thin paste is formed. Ether converts it to a sticky jelly. The secret of dissolving L.V.N. and celloidin (especially the latter) is to wet the material with alcohol before adding the ether. The celloidin should be dissolved separately from the L.V.N. as the presence of the latter prohibits celloidin from dissolving. It is not necessary to stir the mixture until dissolved ; if left overnight all lumps will disappear. Any solution containing

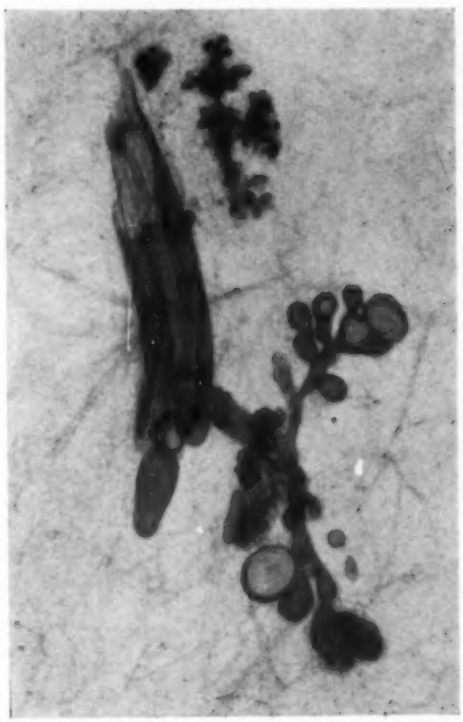

FIG. 266.—Major breast duct with cystic changes in subsidiary branch. (Thick section.) (× 15.)

ether must be kept in a bottle with a ground-glass stopper, evaporation occurring through ordinary stoppers. Solutions containing celloidin or L.V.N. should be kept in wide-necked jars. After using the solution, the ground-glass should be wiped with absolute alcohol to prevent celloidin getting under the stopper.

To make a stock solution :

Quantities :	L.V.N.	140 g.
	Celloidin	4 g.
	Ether	250 c.c.
	Alcohol (Abs.)	210 c.c.

1. Weigh celloidin, and add 50 c.c. of absolute alcohol. Stir to a paste.
2. Add 50 c.c. of ether, stir until the celloidin is dissolved (a few minutes).
3. Weigh the L.V.N. and put into a wide-necked jar. Add the remainder of the alcohol and stir to as near a paste as possible.
4. Add the remainder of the ether, stir to eliminate large lumps.
5. Add celloidin solution to L.V.N.
6. Leave overnight.

This solution contains 20 per cent of L.V.N. and is diluted with equal parts of ether and alcohol as required to constitute the 10 per cent and 5 per cent solutions. Specimens are infiltrated with ether/alcohol mixture followed by 5, 10, and 20 per cent solutions of L.V.N. Impregnation takes place mostly with the 5 and 10 per cent solutions; the 20 per cent solution ensures a block which is hard enough to cut. The following table gives approximate times for embedding but these may be prolonged indefinitely with benefit :—

	Size of Specimen		
	1–2 cm.	*2–5 cm.*	*5–20 cm.*
Ether/Alcohol	1 day	2 days	1 week
5 per cent L.V.N.	1 week	2 weeks	1 month
10 per cent L.V.N.	1 week	3 weeks	1 month
20 per cent L.V.N.	1 week	1 week	2 weeks

Embedding.—The specimen is blocked up in 20 per cent L.V.N. in a paper boat. This is placed in a desiccator with the lid tightly fixed for 24 hours, allowing all the bubbles to rise to the surface. The lid is then left very slightly ajar to allow the ether to evaporate. L.V.N. 20 per cent is used to top up the boats as required. In from 2 to 4 days most small specimens are surrounded by celloidin which has the consistency of soft india-rubber; they are then ready for hardening. Large specimens must be left in the desiccator until they have attained a uniform consistency; this may require a week or more.

Hardening.—The boat containing the specimen is transferred to 70 per cent alcohol for at least two weeks. It may be stored indefinitely in this solution.

Cutting.—The blocks are trimmed with a scalpel, numbered with indian ink and fixed on to the block holder with 20 per cent L.V.N. The block and base are inverted into a shallow dish of 70 per cent alcohol so that the surface of the block is wet, but the 20 per cent L.V.N., acting as glue, is allowed to dry. This may take from ½ to 2 hours.

The block holder must be firmly attached to the microtome, as any movement will result in the block coming off. The knife should cut at an angle of about 45° and make an angle of about 45° with the direction of movement of the blade.

Sections can be cut to any thickness, but it is difficult to obtain them less than 15μ on a big block; to cut thin sections the knife must be very keen. Thick sections are easy to produce and can be cut from blocks up to 5 cm. × 5 cm. on an ordinary sledge microtome. For larger blocks a more rigid instrument is desirable and the Jung 'Tetrander' model has been found excellent for this purpose. After taking a section at 400μ to 500μ, such is the distorting stress upon the knife that it requires adjusting by about a 100μ to be flush with the surface of the block. The block should be damped with 70 per cent alcohol occasionally. Sections are numbered with indian ink; the pen should lightly scratch the nitrocellulose. Each should be numbered on the same surface and in the same corner. Once the ink has partially dried, the section is blotted and stored in 70 per cent alcohol.

Preparation for Staining.—If the specimen has been fixed in a solution containing mercuric chloride the sections should be treated with ½ per cent iodine in 70 per cent alcohol for 24 hours. The iodine is subsequently removed with several changes of 70 per cent alcohol, a process which takes two days or more. For this reason it is best to avoid sublimate fixatives. Formol-fixed sections are washed in distilled water for half an hour, then transferred to 5 per cent aluminium sulphate for 15 minutes.

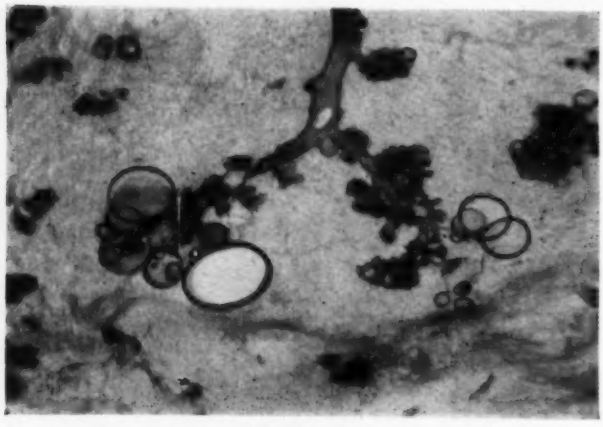

FIG. 267.—Thick sections showing larger terminal cysts. (× 10.)

FIG. 268.—Thick section of a peri-canalicular fibro-adenoma. (× 10.)

Staining.—

1. A stock solution of anthracene blue is made as follows :—

Anthracene Blue (C.I. 1062, G.T. GURR)	0·5 g.
Aluminium Sulphate 5 per cent solution	100·0 c.c.

Boil for twenty minutes, allow to cool, filter next day. For use it is diluted 1–20 to 1–50 times with 5 per cent aluminium sulphate. This is a progressive stain and the weakest strength which is effective is the best. Staining usually takes one to two days. Occasional agitation is necessary in order to obtain uniform staining ; the dye does not penetrate if the section is in contact with the side of the vessel or is touching another section.

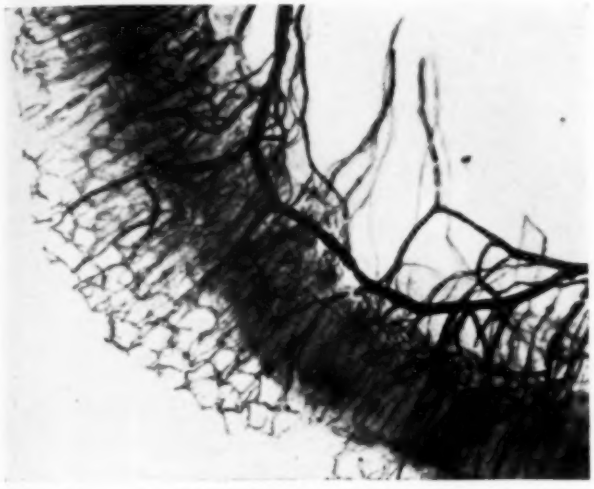

FIG. 269.—Thick section of colon mucosa after injection with indian ink. Notice the honeycomb pattern formed by the capillaries around the necks of glands. (× 20.)

2. Rinse sections in 5 per cent aluminium sulphate.

3. Wash in distilled water for half-an-hour.

4. It is desirable to differentiate the sections ; this frees the celloidin of dye and greatly reduces the intensity of staining of non-epithelial tissues. Epithelial structures are not affected, and by this means excellent contrast is obtained. Differentiation is carried out with 1 per cent aluminium chloride in 90 per cent alcohol and takes from a few hours to several days ; it is difficult to differentiate the sections too much. Differentiated sections are much bluer than those which have not been treated in this way.

5. Wash in distilled water for half-an-hour.

6. Irrigate with running tap water for from 2 to 24 hours.

Mounting.—Sections are treated as follows :—

1. Seventy per cent alcohol for half-an-hour.

2. Ninety per cent alcohol for half-an-hour.

3. Rinse in absolute alcohol. If left for more than a minute in absolute ethyl alcohol, the sections will soften, warp, and finally disintegrate.

4. Absolute isopropyl alcohol for half-an-hour.

5. Transfer to a mixture of 40 parts of cedar wood oil with 60 parts of neutral mounting medium.*

* Obtainable from Messrs. Flatters & Garnett Ltd., 309 Oxford Road, Manchester 13.

This is a useful solution ; it is thin and does not harden nitrocellulose. Sections may be examined in it or stored indefinitely in it. It is best to avoid xylol in the preparation of thick sections as it hardens the L.V.N. and makes it brittle. Cedar wood oil alone makes the sections very brittle.

FIG. 270.—Thick section of stomach mucosa after injection with indian ink. Note two distinct layers of venous drainage. (× 20.)

6. Thick sections need not be mounted at all ; they are conveniently stored in bottles containing the mixture described above. They can also be examined in a Petri dish immersed in the same solution. If it is desired to mount a section, it is left overnight in neutral mounting medium (Flatters and Garnett) which softens the nitrocellulose. It is then placed on a slide with an excess of medium ; as the section is thick, a lot of medium is required to fill in the gap between the cover-slip and slide. The cover-slip is gently pressed on the section between thumb and forefinger of the left hand. This causes most of the bubbles to flow to the edge. Excess of medium overflows the edge and must be removed immediately with a glass pipette connected up to a Sprengel's pump. This method ensures that excess medium does not get on to the cover-slip, hands, etc. If the section has not been softened enough and has some elasticity, it may require constant pressure to flatten it on to the slide. The slide is then left in an incubator overnight, when all the bubbles flow to the edge of the slide. The medium at the edge of the slide hardens rapidly but the remainder stays fluid for many months. Hence it is not wise to trim the sections too soon after preparation ; it is best to get them as clean as possible when first mounted and then leave them for six months. After several months the medium contracts and may cause such stress on the cover-slip that the latter

develops cracks spontaneously. This can be avoided by using a glass-slide instead of a cover-slip.

To Prepare Thin Sections from Thick Ones.—If it is desired to examine the cellular nature of a structure seen in a thick section, this may easily be performed as follows :—

1. Punch or cut out the required segment from the thick section (a paper punch serves very well for this purpose) ; it should be about 0·5 to 0·75 cm. in diameter.

2. Treat with absolute alcohol/ether solution for 24 hours to remove L.V.N.

3. Benzol for half-an-hour.

4. Embed in paraffin wax for 2 hours, but preferably use ester wax which penetrates more quickly at a lower temperature and yields a less brittle block.

5. Continue as for any paraffin section. If the section does not flatten out well in warm water, add about 10 per cent of acetone to the water. Sections are stained routinely ; the anthracene blue already in them can be ignored.

I am very grateful indeed to Mr. E. H. Leach for helping me to modify his original technique and for encouraging me to apply it to surgical problems.

I would like to thank Mr. H. J. B. Atkins for his very kind help and criticism. Mrs. Parnell deserves great credit for her technical skill in preparing the sections.

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SHORT NOTES OF RARE OR OBSCURE CASES

OCCULT CARCINOMA OF THE MALE URETHRA

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"CANCERS of the male urethra are so uncommon that few urologists are experienced in their diagnosis and treatment" (Dean, 1954). Indeed, only 239 proven cases had been recorded before 1951 (McCrae and Furlong, 1951). The following case is of interest because of the difficulty it presented in clinical diagnosis, despite the presence of advanced secondary deposits. The primary growth remained symptomless during life and was not discovered until a post-mortem examination was performed.

CASE REPORT

HISTORY.—The patient was a 67-year-old man who for the previous four years had been a tramp sleeping in open fields and haystacks.

Three months before admission he developed several small, painless swellings in the left groin. These grew slowly and became ulcerated, finally coalescing into one large ulcer.

One month later similar swellings appeared in the right groin and these also became ulcerated.

Two weeks before admission he noticed that his penis and scrotum were swollen. At this time he had slight difficulty in starting micturition, but there was no dysuria or frequency and the urinary stream was good. There was no recent urethral discharge.

In 1916 he had had a urethral discharge treated by irrigations and a second attack of urethritis in 1947, which persisted for several weeks.

He attended the Whitechapel Clinic, London Hospital, on Aug. 20, 1955, having walked from Chelmsford, a distance of 27 miles.

ON EXAMINATION.—On examination he was found to be a thin, unkempt, elderly man with extensive ulceration in both groins, more marked on the left side (*Fig.* 271). The largest ulcer measured 10 × 6 × 2 cm. deep, was irregular in shape, with raised edges and surrounding induration. The floor was covered with a thick yellow slough and beneath this the left femoral artery was beating ominously. There were several smaller similar ulcers on the right side. In the surrounding skin and that of the penis and scrotum there were scattered white, hard nodules up to 0·8 cm. in diameter. The penis and scrotum were grossly œdematous and there was a tight phimosis. The glans penis could not be felt. No tumour was found on the legs or buttocks, or on rectal examination, and, except for a smooth enlargement of the liver, no further abnormality was found on routine physical examination. His urine showed a few threads in the first glass of the two-glass test, but was otherwise normal.

Investigations.—Blood Wassermann, Price's precipitation reaction, and gonococcal complement-fixation tests were negative. Frei and Dmelcos intradermal tests were also negative, as was a complement-fixation test for lymphogranuloma venereum. Biopsy of the ulcers from both groins showed squamous-cell carcinoma.

A dorsal slit was performed and revealed the glans penis to be normal. Urethral bougies to 14/16 Charrière were passed without difficulty and examination with the bougies in situ revealed no abnormality on perineal or rectal palpation. Sigmoidoscopy was normal to 18 cm.

X-ray therapy, with a total dose of 3,500 r to each groin in divided doses, was given daily for 15 days, but produced no improvement.

The patient became drowsy, his condition slowly deteriorated, and he died on Sept. 30.

POST-MORTEM EXAMINATION.—There was an ulcerating carcinoma 3 cm. long, incompletely encircling the penile urethra 3 cm. distal to the verumontanum, with direct invasion of the erectile tissue around the growth and four secondary deposits in the corpora cavernosa (*Fig.* 272). *Fig.* 273 shows the ulcerating nature of the lesion. Apart

from some congenital cysts in the left kidney, the rest of the urinary tract was normal. Multiple secondaries were found in the lower half of the right rectus abdominis and

FIG. 271.—The condition on admission, showing the ulceration in the groins and the subcutaneous nodules in the penis.

FIG. 272.—The bladder and urethra opened to display the primary tumour.

multiple osteolytic secondaries in the first and fifth lumbar vertebræ. Scanty secondary plaques were present in the peritoneum and both pleuræ showed many secondary nodules. These were also present in the right hilar group of lymph-nodes. The iliac glands were filled with secondary deposits, but the para-aortic glands were clear. There were two capsular secondaries on the anterior surface of the right lobe of the liver.

Histological Examination.—This showed rather poorly-differentiated squamous carcinoma of the urethra, with only occasional cell nests and small foci of keratinization.

COMMENT

Ulcerating carcinoma of the urethra is uncommon, the usual type being a ring carcinoma, associated in 48 per cent of cases with a pre-existing stricture (McCrae and Furlong, 1951). As a result, the correct diagnosis is usually not made before the onset of obstruction to the urinary flow, although in the penile urethra, tumour, fistula, or urethral discharge may lead to an earlier diagnosis.

Histologically, squamous-cell carcinoma is the most common type of tumour, accounting for 69 per cent of cases. This high incidence in a structure normally lined with transitional-cell epithelium may be due to metaplasia secondary to chronic irritation or to the normal presence of islands of squamous epithelium.

Many patients, as in this case, are stated to be of poor mentality and low social status, which may account for the delay in seeking medical advice.

SUMMARY

1. A case of occult carcinoma of the male urethra is described.

2. The clinical and pathological features of the disease are briefly considered.

FIG. 273.—Close-up view of the urethra, showing the ulcerated nature of the tumour.

We should like to thank Mr. Ambrose King for permission to record this case; and Professor Dorothy Russell for the pathological reports.

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PRIMARY CARCINOMA OF THE NAIL-BED

By B. S. ASHBY

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THE nail-bed is an unusual site for malignant disease. Both malignant melanoma and squamous carcinoma are described. Of the latter, 25 examples have been traced in the literature since 1850, and the earliest description to appear in the British journals was by Jonathan Hutchinson (1885).

CASE REPORTS

Case 1.—Mrs. E. W., aged 78 years (Case No. R 9873), was admitted to Westminster Hospital on Sept. 20, 1955, complaining of pain and swelling in the distal phalanx of the left index finger for seven years. There was no antecedent trauma, and she had never had any occupation other than housework. She was right-handed. She had attended a number of hospitals for treatment; the nail had been removed on five occasions, the last being fourteen months ago, and courses of penicillin and ultra-violet light had been given without effect.

FIG. 274.—*Case* 1. Pre-operative clinical photograph of left index finger.

The pain and swelling progressively increased in spite of treatment, and she first presented in the dermatological department of this hospital one year ago with the clinical appearances of a severe chronic paronychia. A course of X-ray therapy was given with no effect, and she was admitted for amputation.

On examination she was an essentially normal, active old lady. The terminal phalanx of the left index finger was symmetrically swollen, reddened, and very painful; the nail was thickened, elongated, and discoloured (*Fig.* 274); there was marked tenderness such that she guarded the finger. There were no palpable lymph-nodes in the areas of regional drainage. A diagnosis of chronic paronychia was made. X-ray examination revealed some osteoporosis of the phalanges, but no bone erosion or destruction.

Amputation was carried out through the proximal interphalangeal joint under general anæsthesia, with uneventful recovery. The histology report was:—"The epidermis of the nail-bed is thickened by a small basi-squamous carcinoma" (*Fig.* 275).

At the time of writing, after nine months, the patient is alive and well.

Case 2.—Miss E. T., aged 53 years (Case No. U 5184), a physiotherapist, was admitted on June 19, 1956. She was right-handed. Five years previously she had injured her right index finger in a car door. Since then the nail had been slightly deformed, and at its ulnar border a skin crack appeared and healed again on several occasions. Six months ago such a crack occurred but failed to heal, developing into a pale wart-like lesion spreading beneath the nail, the ulnar half of which became yellowish-white in colour. The finger was painless, and had never bled. She was first seen at another hospital where a biopsy was taken. The histology report was: "Squamous-cell carcinoma showing moderate keratinization."

She was referred to this hospital, and the finger was amputated through the metacarpophalangeal joint under general anæsthesia, with uneventful recovery. The histology report was negative, but in view of the biopsy findings a diagnosis of carcinoma of the nail-bed was made.

FIG. 275.—*Case* 1. Low-power view of nail-bed showing carcinomatous invasion. Hæmatoxylin and eosin. (× 6·6.)

There was no glandular enlargement up to the time of biopsy. When seen at Westminster Hospital the epitrochlear gland was enlarged and tender, but as the biopsy site had become infected, and the enlargement disappeared rapidly post-operatively, it was regarded as inflammatory.

DISCUSSION

Case 1 presents a number of unusual features, making it of especial interest, and in some respects unique, amongst the 25 examples of carcinoma of the nail-bed in the literature. Levine and Lisa (1939) reviewed 18 cases reported up to that time, and described another occurring in association with a left ingrowing great toe-nail. Those described since that paper are listed in *Table I*.

The incidence of the condition is almost equally divided between the sexes, there being 14 male and 10 female recorded, with 1 unspecified; both the present patients are female. The age distribution is wide, between 20 and 75 years. The majority occur in the hand, the commonest being the right thumb and right index finger. The site distribution is summarized in *Table II*. It will be seen that *Case* 1 is the first example in which the condition has occurred in the left hand.

Most examples appear to be associated with injury, or injury followed by chronic infection.

Silverman (1935) reports a case following one year's paronychia after a puncture injury, in which the nail was removed three times. Some follow chronic infection associated with ingrowing toe-nails. Malignant change in a simple chronic paronychia, with absence of trauma, is unusual. No history of trauma was obtained from the patient in *Case* 1, but during the seven years' chronic infection the nail was removed five times. Instances are recorded of other antecedent conditions, such as Bowen's disease, and following the hyperkeratosis of arsenic poisoning (Pardo-Castello, 1941).

Table I.—List of Cases described since the Last Review in 1939

	Sex	Age	Site
Pardo-Castello, 1941 (also mentions 2 others first described in 1936)	Male	45	Right middle finger
Ellis, 1948	Male	73	Right thumb
	Male	66	Thumb
	Female	70	Right middle finger
Russell, 1950	Male	37	Right index finger
John, 1956	Female	60	Right thumb

The histological structure in most of the reported cases was of squamous carcinoma, though 2 were basal-cell type. Histology of the tumour in *Case* 1 showed a basi-squamous carcinoma, areas of basal-cell masses alternating closely with epithelial whorls (*Fig.* 276). *Case* 2 was a typical squamous-cell carcinoma.

Carcinoma of the nail-bed, as far as can be judged, is of low-grade malignancy (Bunnell, 1944; Russell, 1950). Lymph-node metastases are rare, and in most instances a curative radical amputation has been performed. Erosion and destruction of the underlying bone are known, but unusual, 2 cases of the former and 5 of the latter having been reported (Ellis, 1948; Russell, 1950).

SUMMARY

Two examples of primary carcinoma of the nail-bed are described. *Case* 1 is the first recorded in the left hand. The cases reported since the last review are listed. Site, aetiology, and pathology are briefly discussed.

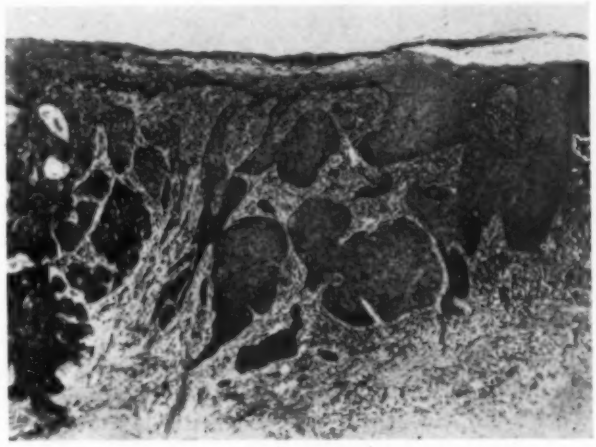

Fig. 276.—*Case* 1. Higher magnification to show basi-squamous characters. Hæmatoxylin and eosin. (× 26·6.)

Table II.—Site Distribution of All Recorded Cases of Carcinoma of the Nail-bed

		1st Digit	2nd Digit	3rd Digit	4th Digit	5th Digit	Total	
Hand	Right	8	4	2	—	—	14	
	Left	—	—	—	—	—	—	19
	Unspecified	3	—	2	—	—	5	
Foot	Right	1	—	—	—	1	2	
	Left	2	—	—	—	—	2	6
	Unspecified	2	—	—	—	—	2	
							Total	25

I would like to thank Mr. E. Stanley Lee and Dr. Peter D. Samman for permission to publish these cases; also Dr. A. D. Morgan for advising upon and making the photomicrographs; and Dr. Peter Hansell for permission to publish the clinical photograph.

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REVIEWS AND NOTICES OF BOOKS

Surgery of the Eye: Diseases. By ALSTON CALLAHAN, B.A., M.S., M.D., F.R.C.S., Birmingham, Ala. 11 × 8½ in. Pp. 448 + xxvi, with 263 illustrations. 1956. Springfield, Ill.: Charles C. Thomas. (Oxford: Blackwell Scientific Publications.) £9.

THIS is a companion volume to the author's first book *Surgery of the Eye: Injuries*, and it is written in the same excellent style and produced in the same format.

The majority of the operations are presented in steps arranged on a single page, often laid out opposite the explanatory text. This procedure eliminates explanatory titles of the illustrations which would require a second caption.

One of the features of this large work is the excellence of the illustrations, many of which are original.

The section dealing with corneal opacities and corneal grafting is a model of completeness and could not be bettered.

An excellent production which will appeal to all ophthalmic surgeons on both sides of the Atlantic.

A Manual of Anæsthetic Techniques. By WILLIAM J. PRYOR, F.F.A.R.C.S. (Eng.), M.F.A., R.A.C.S., Anæsthetist, Thoracic Unit, Christchurch Hospital, New Zealand. 8½ × 5½ in. Pp. 212 + ix, with 77 illustrations. 1956. Bristol: John Wright & Sons Ltd. 27s. 6d.

THIS book, we are told, is written primarily for House Surgeons and Registrars commencing the practice of anæsthetics, but it is also claimed as an attempt to include in one cover most of the important changes in recent years. These two aims are scarcely compatible, and the second would need a larger volume than this for its fulfilment. In consequence, some of the material—such as a description of methods of anæsthesia for laryngectomy, lobectomy, and Blalock's operation—is beyond the scope of the novice; and the rest is on too elementary a plane for the expert.

With this exception the book may be commended to its intended readers. It is clear, simple, factual, and direct, and contains much useful information. It is wide in scope, and includes chapters on premedication, intravenous therapy, anæsthetic emergencies, local anæsthesia, dental anæsthesia, and post-operative care.

Though the expert may find it a little unscientific in its approach, there are few books available at present which present the subject to the novice with such clarity and good sense.

Urological Surgery. By AUSTIN INGRAM DODSON, M.D., F.A.C.S., Professor of Urology, Medical College of Virginia; etc. With nine contributors. Third edition. 9¾ × 6¾ in. Pp. 868, with 664 illustrations. 1956. London: Henry Kimpton. £7 10s.

THE third edition of this comprehensive volume on urological surgery has been completely revised so as to present the present trends in this branch of surgery.

On the whole British urologists will find that nephroptosis is given too much space in a work of this calibre. Twenty pages are devoted to this subject and five different operations dealing with nephropexy are described.

Perineal prostatectomy is described in detail and some twenty pages are devoted to this operation, which is very rarely performed to-day.

This volume is well illustrated and gives a good account of urological surgery as practised in America.

Sir John Bland-Sutton, 1855–1936. By W. R. BETT, M.R.C.S. (Eng.). With a Foreword by Lord WEBB-JOHNSON. 8¾ × 6½ in. Pp. 100 + viii, with 6 Plates. 1956. Edinburgh: E. & S. Livingstone Ltd. 20s.

THIS is an excellent short history of Sir John Bland-Sutton, who was President of the Royal College of Surgeons of England from 1923 to 1925. Sir John had many facets to his character, and was a great student of nature and the many and various papers from his facile pen always had a background of anatomy, embryology, or morphology. He enriched the Hunterian Museum of the College of Surgeons not only with pathological specimens but also with comparative anatomy specimens from the London Zoo and from countries overseas.

We very much hope that some day a larger volume will be published dealing with the versatile personality and works of Sir John Bland-Sutton. Dr. Bett has only whetted our appetites by his admirable little book.

Histological Appearances of Tumours. With a Consideration of Their Histogenesis and Certain Aspects of Their Clinical Features and Behaviour. By R. WINSTON EVANS, T.D., B.Sc., M.R.C.S., L.R.C.P., Consultant Pathologist, United Liverpool Hospitals; Lecturer in Clinical Pathology, University of Liverpool; etc. 9¾ × 7¼ in. Pp. 773 + xvi, with 980 illustrations. 1956. Edinburgh: E. & S. Livingstone Ltd. 90s.

THIS is an excellent book which will make a strong appeal to all surgeons who are interested in the tumour pathology of their patients.

Not only are the histological appearances of tumours considered and illustrated in detail, but the author also relates their histogenesis and portrays certain aspects of their clinical features and behaviour.

There are nearly one thousand illustrations, all of which are clear and beautifully reproduced. The histological appearances of all human tumours are to be found in this work, and the magnification of every photomicrograph is given together with a note of the staining process.

As a reference volume for surgeons and pathologists this book could not be bettered; and it should prove of real value to the younger generation of surgeons who are working for higher surgical examinations.

At the end of each chapter a comprehensive list of modern references dealing with the particular subject is given; and these references include some from the other side of the Atlantic.

An excellent production and a most useful book.

A Search for Similarities. The Ninth Macewen Memorial Lecture delivered in the University of Glasgow, April 15, 1955. By Sir JAMES LEARMONTH, K.C.V.O., C.B.E., Ch.M., LL.D., F.R.C.S.E., Regius Professor of Clinical Surgery and Professor of Surgery, University of Edinburgh. 8½ × 5½ in. Pp. 34. 1956. Glasgow: Jackson, Son & Co. 5s.

FELLOWS of the Association of Surgeons who heard Sir James Learmonth deliver this ninth Macewen Memorial Lecture will be glad to have it to read; and surgeons who were not present on that occasion, particularly those who are teachers and any who are specially interested in the vascular system, will wish not only to read it now but also to add it to their libraries. It is of interest as a model of the philosophic approach and the breadth of view which such a Memorial Lecture demands, but it is of special importance because it describes the lecturer's

personal experience of peripheral vascular disorders in childhood, and will therefore prove to be of great value to anyone faced with one of these rare and obscure conditions.

Sir James begins with the congenital lesions, the single and multiple arteriovenous fistulæ, and the even less common congenital malformations of lymphatics. He then describes the anatomical variations from the normal pattern of arteries and veins, before reviewing the pathological conditions affecting the vascular system in childhood, seeking for the similarities and differences between these and the vascular diseases of adult life. It is a masterly survey, an arresting feature of which is the stress laid upon venous thrombosis as a cause of nutritional lesions varying in extent from small cutaneous losses to massive gangrene. When venous thrombosis produces gangrene the local arteries are patent up to the line of demarcation, and a remarkable feature is that the swollen, cyanosed part does not feel icy cold as do parts deprived of their arterial supply.

The lecture is both a source of information and a stimulus to further study, and is a worthy tribute to the memory of Sir William Macewen.

Principles of Chest X-ray Diagnosis. By GEORGE SIMON, M.D., F.F.R., Assistant Director, X-ray Department, Brompton Hospital, London; Radiologist, St. Bartholomew's Hospital, London; etc. 11 × 8½ in. Pp. 183 + x, with 162 illustrations. 1956. London: Butterworth & Co. (Publishers) Ltd. 50s.

THIS is a unique book in many ways, for the author has adopted a new approach to the subject of chest radiographs. The material in the book has been arranged under headings, rather than the usual clinical disease labels.

In such a book as this the pictorial side is all-important and Dr. Simon certainly has collected together over one hundred and fifty excellent radiographs of the chest, all of which have been most carefully chosen to depict a definite condition.

This book should prove a boon to all who are interested in chest diseases.

The Year Book of Urology, 1955–1956. Edited by WILLIAM WALLACE SCOTT, M.D., Ph.D., Director, James Buchanan Brady Urological Institute, The Johns Hopkins Hospital; etc. 7⅝ × 5⅛ in. Pp. 398, with 82 illustrations. 1956. Chicago: Year Book Publishers Inc. (London: Interscience Publishers Ltd.) 49s.

YEAR by year as the art of Urology progresses the value of this book increases. This volume contains an admirable summary by the editor of the proceedings of the tenth congress of the International Society of Urology. For the rest it follows the usual pattern of giving abstracts of current literature sufficiently long to whet the appetite of the reader to consult the original articles. Used in this way and not as a substitute for reference to the original, it is a very great aid to the serious student of the subject.

Skin Surgery. By ERVIN EPSTEIN, M.D., Assistant Clinical Professor of Medicine (Dermatology), Stanford University Medical School; etc. 9¼ × 6¼ in. Pp. 228, with 101 illustrations. 1956. London: Henry Kimpton. 56s.

THE title of this monograph is far from accurate, for one can hardly include the surgery of mixed palatine tumours, cancers of the mouth, lip, jaws, and penis as coming under the purview of a dermatologist. It is true that Dr. Epstein has obtained the services of other medical men to write some of the chapters in this book. If, as is stated in this book, "Dermatologic Surgery is usually an office procedure, carried out in clean but not completely 'aseptic' fashion", why describe and illustrate surgical procedures which rightly belong to the domain of general surgery and are major operative procedures?

The best chapters in this book are concerned with the value of biopsy and the technique of cautery excision.

The Dental Treatment of Maxillo-facial Injuries. By Sir WILLIAM KELSEY FRY, C.B.E., M.C., F.R.C.S., F.D.S., Consulting Dental Surgeon to the Royal Air Force, the Ministry of Health, and the Institute of Dental Surgery; Consulting Dental Surgeon Emeritus to Guy's Hospital; and TERENCE WARD, M.B.E., F.D.S.R.C.S. (Eng.), Consulting Dental Surgeon to the British Army and to the Queen Victoria Hospital, East Grinstead. Second edition. 8¾ × 5½ in. Pp. 372 + xii, with 383 illustrations. 1956. Oxford: Blackwell Scientific Publications. 47s. 6d.

THE second edition of this classic work will prove as popular as the first edition, which was published during the second world war and was reprinted three times.

The new edition has been thoroughly revised and brought up to date and Sir Archibald McIndoe has contributed an excellent chapter on "Fractures of the Middle Third of the Face" which enhances the value of the book.

We have nothing but praise for this book, for it portrays the very best of British Dental Surgery; it is an excellent production.

A Manual of Fractures and Dislocations. By BARBARA BARTLETT STIMSON, A.B., M.D., Med. Sc. D., F.A.C.S., Director of Department of Bone and Joint Surgery, St. Francis Hospital, Poughkeepsie, New York. Third edition. 7⅝ × 5⅛ in. Pp. 224, with 97 illustrations. 1956. London: Henry Kimpton. 34s.

THIS small book is described as being primarily written for medical students. As such it forms a useful contribution to their library. The arrangement of the material makes it very easy to read, and particularly useful to the student are the summaries at the end of each section. It is good to see the emphasis put on the clinical examination of the injuries rather than on X-rays.

An orthopædic surgeon is bound to find differences of opinion in the text; for instance, one would question the use of Bryant's traction for fractures of the femur in children up to five years of age, particularly in view of the possible vascular complications. It is interesting to see that the reduction and immobilization of simple crush fractures of the spine is still advised. However, these are minor criticisms and do not in any way reduce the value of the work.

Tumours of the Major Salivary Glands. By FRANK W. FOOTE, jun., M.D., and EDGAR L. FRAZELL, M.D. Pp. 149. 1956. Washington, D.C.: Armed Forces Institute of Pathology. $1.50.

THIS is section IV—Fascicle II of the *Atlas of Tumour Pathology* which is well known the world over. This excellent section is the outcome of a careful study of some 900 cases seen at the Memorial Hospital during a 20-year period ending in 1949. Never before has such a large series of tumours been submitted to such a searching pathological study.

There are some 180 pictures in this monograph, the majority of which are photomicrographs of truly excellent quality, all of which are reproduced on art paper. These are some of the best photomicrographs we have ever seen,

and should prove of real value not only to pathologists but also to those surgeons who are interested in the pathological examinations of tumours they have excised.

This section is of the same high-class production as the former published sections and we congratulate the Armed Forces Institute of Pathology on an excellent job of work.

Campbell's Operative Orthopædics. Edited by J. S. SPEEL, M.D., assisted by ROBERT A. KNIGHT, M.D. Third edition. In two volumes. 9¾ × 6¾ in. Vol. I, Pp. 1104 + xxii, with 68-pp. Index to vols. I and II; Vol. II, Pp. 1105–2124 + xvi, with 68-pp. Index to vols. I and II. 1956. London: Henry Kimpton. £14 10s.

THE third edition of this comprehensive work on operative orthopædics is mainly due to the combined efforts on the part of some sixteen authors, all of whom are members of the staff of the Campbell Clinic and Faculty of the University of Tennessee College of Medicine, Memphis.

Every branch of orthopædic surgery is thoroughly covered and well illustrated with many original pictures.

In this traumatic age, as one would expect, a large part of this work is devoted to the treatment of fractures. American treatment of fractures is inclined to be far more operative than it is over here. Medullary fixation of fractures by a Rush pin is rarely undertaken in England.

The section on amputations is very complete and practical and beautifully illustrated. Gordon-Taylor's hindquarter amputation is well described.

The subject of arthroplasty receives due attention and the many and varied operative techniques are well illustrated.

These two large volumes will still remain the universal reference work on operative orthopædics.

The Royal Northern Hospital, 1856–1956. By ERIC C. O. JEWESBURY, M.A., D.M. (Oxon.), M.R.C.P. With a Foreword by H.R.H. The DUKE OF GLOUCESTER, K.G. 8½ × 5¼ in. Pp. 157 + xii, with 23 plates and 5 illustrations. 1956. London: H. K. Lewis & Co. Ltd. 17s. 6d.

IT may be said that the Royal Northern Hospital owes its origin to a young surgeon who while on the Staff of University College Hospital playfully slapped the buttocks of a man on the operating table. His name was Sherard Freeman Statham and it was none other than John Erickson who was responsible for his dismissal. It was on May 28, 1856, that Statham ceased to be Assistant Surgeon to University College Hospital.

However, Statham was a man of enterprise and he was determined to continue to practise surgery. He, therefore, secured the lease of a property in York Road, King's Cross, and with the help of his brother, who was a dentist, and Dr. Whidborne and Dr. Croft he founded the Great Northern Hospital with some sixteen beds. All this happened a hundred years ago and since that time the hospital has gone from strength to strength.

Dr. E. C. O. Jewesbury has presented the medical profession with an excellent history of this hospital, and from it can be gleaned the many 'ups' and 'downs' the hospital suffered in a century.

Chirurgie Réparatrice. By MERLE D'AUBIGNÉ. 11 × 7 in. Pp. 268, with 178 illustrations. 1955. Paris: Expansion Scientifique Française. 3,900 fr.

PROFESSOR D'AUBIGNÉ, who is as well known and respected in Britain as he is in France, has sought the help of twelve of his colleagues and assistants at the Cochin Hospital, Paris, to study a limited number of important orthopædic problems. The result of this study is now published in a kind of 'recent advances' series. The book is thus a collection of unrelated chapters, each written by one or two surgeons of the Orthopædic Department of the Cochin Hospital. The subjects discussed are: the functional results of hip arthroplasty; damage to the ligaments and cartilages of the knee; non-union of the tibia; arthrodesis of the ankle; tabetic arthropathy; paralysis of the brachial plexus; malunion of the lower end of the radius; non-union of the carpal scaphoid; Dupuytren's contracture; restoration of opposition to the thumb; treatment of ulnar claw-hand; myelography; and burns. Each chapter is completed by a very short summary in English.

The bibliography is adequate; the illustrations, photographs, and radiographs leave nothing to be desired; and altogether the book is very well produced, and contains much of interest to British surgeons.

Atlas de Chirurgie Thyroïdienne. By JACQUES FRANCILLON and CLAUDE GABRIELLE, Lyons. With a Foreword by LÉON BÉRARD. 10⅝ × 7½ in. Pp. 182 + vi, with illustrations by Dr. C. MILLET. 1955. Paris: Expansion Scientifique Française.

PROFESSOR LÉON BÉRARD, in the foreword, states that the book is intended to help and guide those general surgeons who want to improve their technique in the surgery of the thyroid gland. The book is an analysis of the successive stages of thyroidectomy, and is very freely illustrated. Details of the difficulties and dangers sometimes encountered are given particular emphasis; for instance, 28 pages are devoted to the variations in the anatomy of the recurrent laryngeal nerves and the parathyroids.

Throughout the book each step of the operation of thyroidectomy is explained in clear-cut sentences and illustrated by an appropriate diagram. The authors appear to favour a U-shaped skin incision, but discuss and illustrate alternative incisions. Although the book contains nothing new, there is much in it that may prove helpful to many young surgeons.

Gynecologic Cancer. By JAMES A. CORSCADEN, M.D., Emeritus Professor of Clinical Gynecology, College of Physicians and Surgeons, Columbia University. Second edition. Pp. 546 + x, with 115 illustrations. 1956. London: Baillière, Tindall & Cox Ltd. 80s.

CANCER of the female genital tract is still all too common, and if better results from operation are to ensue then earlier diagnosis must be possible. This is the theme all through this book. During the last decade the treatment of cancer of the cervix by radiotherapy has become more stabilized on both sides of the Atlantic, and Dr. Corscaden has given an excellent account of the radiotherapy 'set-up' in America.

It is interesting to note that in New York City the Papanicolaou smear for vaginal exfoliative cytology is now available to any female resident regardless of her economic status.

This book gives an excellent account of the current American views on the diagnosis and treatment of cancer of the female genital tract.

Diagnosis and Treatment of Vascular Disorders (Angiology). Edited by SAUL S. SAMUELS, A.M., M.D., F.A.C.A., F.A.C.C., Editor-in-Chief, Angiology; President, Angiology Research Foundation; etc. 10 × 6⅞ in. Pp. 621 + x, with numerous illustrations. 1956. London: Baillière, Tindall & Cox Ltd. 128s.

THIS book has been written by seventeen contributors, including four—Boyd, Goetz, Ratcliffe, and Rose—who practise in the British Commonwealth and who have written one-third of the contents. Under the editorship

of Saul S. Samuels, it has been well produced and contains much that is of value, but there are certain notable omissions, particularly that of lymphangiography from the chapter on angiography and of any adequate description of the syndrome of thrombosis of the distal abdominal aorta and iliac arteries.

The chapters by R. H. Goetz on examination of the patient and on Raynaud's phenomenon, by F. A. Simeone on arterial injuries, and Saul S. Samuels on thromboangiitis obliterans are outstanding. The book is recommended to those interested in the diagnosis and treatment of vascular disorders.

Pulmonary Carcinoma. Pathogenesis, Diagnosis, and Treatment. Edited by EDGAR MAYER, M.D., and HERBERT C. MAIER, M.D. 9 × 6 in. Pp. 540 + xvi, with numerous illustrations. 1956. New York: New York University Press. (London: Pitman Medical Publishing Co. Ltd.) £6.

THIS large volume is devoted to the pathogenesis, diagnosis, and treatment of cancer of the lung, one of the commonest forms of malignant disease met with to-day.

The authors have collected together seventeen contributors, each one writing on his own particular sphere; so much so that every conceivable facet of cancer of the lung is covered in a most comprehensive manner.

No one better qualified than Dr. Cornelius Rhoads could write on the modern concepts of cancer research; with his extensive background at the Memorial Centre in New York he has probably a better 'bird's-eye' view of the future of this problem than anyone else.

There can be no doubt whatever that cancer of the lung is a health problem of alarming proportions both in England and America, and it therefore behoves the medical profession as a whole to be *au fait* with the early signs and symptoms of the disease.

This volume includes excellent practical chapters on the surgical technique of pneumonectomy, radiotherapy, chemotherapy, the use of isotopes, the management of inoperable cases, and the overall management of patients with malignant disease of the lung.

This is a comprehensive work which reflects the opinion of American physicians and surgeons on the treatment of an all-too-common disease.

The book is profusely illustrated but some of the radiographs are not very good.

The Pathology and Surgery of the Veins of the Lower Limbs. By HAROLD DODD, Ch.M., F.R.C.S., Surgeon to St. Mary's Hospital, etc.; and FRANK B. COCKETT, M.S., F.R.C.S., Surgeon to St. Thomas's Hospital. With a Foreword by R. R. LINTON, M.D., Assistant Clinical Professor of Surgery, Harvard Medical School. 9¾ × 6¾ in. Pp. 462 + xii, with 313 illustrations. 1956. Edinburgh: E. & S. Livingstone Ltd. 65s.

IN under 500 pages the authors have provided most of the information available and all that is necessary about the pathology and surgery of the veins of the lower limb. A considerable part of the book is devoted to what one might term 'the basic sciences', and rightly so, because it is upon a proper understanding of these that correct diagnosis and treatment is based. Particular insistence is placed upon a precise diagnosis of the type and content of varicose veins and the correct placing of the incompetent valves upon which these depend. The various tourniquet tests are described in considerable detail and the value of the information which these impart is fully analysed. In discussing the treatment of varicose veins, the dwindling place which injection treatment holds and the indications for its use are well described. The habit of injecting veins with the patient standing is condemned and the technique of needle insertion is described, although more faith is placed in the complete transfixion and withdrawal technique than most experienced surgeons would allow. As for operative treatment, although other methods of saphenectomy in situ (with the exception of the occasionally useful diathermy coagulation) are mentioned, the authors are convinced of the paramount place occupied by the stripping operation and impart their conviction to the reader. The technique is well illustrated and the young surgeon is given excellent advice throughout on how to perform this common, although occasionally hazardous, procedure. In a highly specialized book of this nature it might, however, be considered somewhat superfluous to depict how to sew up the skin with eversion of the edges and to caution against overlap.

The chapter on thrombosis and embolism is excellent and the advice as to treatment entirely acceptable. It is not, however, apparent why heparin should be prescribed in those cases presenting as a pyrexia of unknown origin where the diagnosis may be in doubt, but should be withheld from those cases presenting with local signs; perhaps a later edition will make the rationale of this view more clear. Venography (with suitable apologies for the etymological solecism) is indicated in very few cases, but the technique of its performance together with the difficulties of interpretation are described.

In recurring embolism, venous ligation is advised. Where the source of the embolus is thought to be within the pelvis, bilateral common iliac ligation is preferred to ligation of the inferior vena cava because some collateral circulation is thereby preserved. When the source of the embolus is from the legs, femoral ligation distal to the profunda tributary is advocated. The book ends with a very good account of the pathology, diagnosis, differential diagnosis, and treatment of nutritional ulcers. This is essentially a work which the practising surgeon should possess, the 'Fellowship candidate' peruse, and of which the publishers may as usual be justly proud.

Studies in Topectomy. Edited by NOLAN D. C. LEWIS, M.D., CARNEY LANDIS, Ph.D., D.Sc., and H. E. KING, Ph.D. 9 × 6 in. Pp. 248 + viii. Illustrated. 1956. New York and London: Grune & Stratton. $6.75.

THIS monograph on topectomy is made up of many contributions from numerous psychiatrists and psychologists and psychosurgeons. Topectomy is the operative excision of the frontal and orbital cortex of the brain. These areas of the brain are exposed through an osteoplastic flap and the cortex is removed to a depth of 2 cm. Careful hæmostasis is carried out with electrocautery and gelfoam. Few complications follow this operation and patients have been investigated clinically over a long period.

In some of the psychiatric disorders this operation seems to be superior to that of lobotomy.

Clinical Urology. By OSWALD SWINNEY LOWSLEY, A.B., M.D., F.A.C.S., F.I.C.S., New York; and THOMAS JOSEPH KIRWIN, M.A., M.S., M.D., F.A.C.S., F.I.C.S., New York. Third edition. 11 × 8½ in. In 2 volumes. Pp. 985 + xiv, plus complete Index in each volume. 1956. Baltimore: The Williams & Wilkins Co. (London: Baillière, Tindall & Cox Ltd.) £12 12s.

IT is twelve years since the second edition of this extensive work was published, but the edition was reprinted no less than five times. This in itself indicates the popularity this comprehensive book enjoys.

The third edition has been carefully revised and much of the text has been rewritten. The pages have been rearranged into three columns and enlarged to make the text easier for reading.

One of the features of these two large volumes is the excellence of the illustrations throughout; the majority of these come from the pen and brush of the well-known urological artist, William P. Didusch. The excellence of these illustrations makes this work an atlas of urological pathology and surgical technique.

There can be little doubt that as a work of reference this work is unsurpassed; and it should prove of real value to the graduate who wishes to specialize in urology.

Modern Gynæcology with Obstetrics for Nurses. By W. E. HECTOR, Principal Tutor, St. Bartholomew's Hospital, London; and JOHN HOWKINS, M.D., F.R.C.S., Assistant Gynæcologist and Obstetric Surgeon, St. Bartholomew's Hospital, London. 8½ × 5½ in. Pp. 275 + viii, with 99 illustrations. 1956. London: William Heinemann. 17s. 6d.

THIS is a well-illustrated and clearly printed book written by the Principal Tutor, in conjunction with one of the Gynæcologists at St. Bartholomew's Hospital, as a guide to nurses who are about to undertake the special nursing care of obstetrical and gynæcological patients. A wide field is covered, including not only the anatomy and physiology of reproduction but also the pathology of the female organs of generation and an account of the commoner obstetrical and gynæcological operations. The line drawings throughout the text are plentiful and excellent. Among the instruments listed or illustrated, some specialism may be detected in the mention of Maingot's hysterectomy clamps and McIndoe's scissors, while Berkeley's and Bonney's names are conspicuously absent; but such, along with the absence of any mention of the Higginson's syringe, which was at one time almost the badge of office of the midwife, may be signs of the times.

The book can be confidently recommended to those for whom it is written.

The Life of Hugh Owen Thomas. By DAVID LE VAY, M.S. (Lond.), F.R.C.S. (Eng.), Orthopædic Surgeon, S.E. Metropolitan Region. 8¾ × 6¼ in. Pp. 136 + viii, with 14 plates and 13 illustrations. 1956. Edinburgh: E. & S. Livingstone Ltd. 25s.

MUCH has been written about Hugh Owen Thomas, but there is certainly room for this excellent small volume because it gives the reader a concise and intimate account of his life.

Mr. Vay is to be congratulated on the way he has marshalled his facts regarding Thomas and the way he has presented them. Few surgeons to-day have any idea of the enormous practice and the variety of surgical work that was undertaken by Thomas.

The illustrations throughout the book are excellent and are beautifully reproduced.

This monograph will take its place among the many which have appeared from the House of Livingstone during the present decade.

The Organization of the Cerebral Cortex. By D. A. SCHOLL, Reader in Anatomy, University of London. 8½ × 5½ in. Pp. 125 + xvi, with 12 plates and 11 diagrams. 1956. London: Methuen & Co. Ltd. (New York: John Wiley & Sons Inc.) 18s.

THE author quotes P. A. M. Dirac, that "the main object of physical science is not the provision of pictures, but is the formulation of laws governing phenomena and the application of these laws to the discovery of new phenomena", and then sets out to review our knowledge of the cerebral cortex and attempts to explain its working according to a mechanistic view. This is based on a mathematical conception of cortical organization. Only in a few areas of the cerebral cortex is it possible to make fairly sharp regional demarcations; with few exceptions, transition from one region to another is gradual and we are given a salutary reminder of the variations in the maps of the cortex made by different observers. It is possible to describe the dendritic fields of the cortical neurons quantitatively and this book is a plea for a statistical approach to any study of cortical architecture. It may be argued that such a mechanistic view overlooks the qualitative at the expense of the quantitative and that no increase in quantity can correct defects in quality. It is a far cry from a digital computer to the production of *Paradise Lost*. There is a brief bibliography of this vast subject which has been attractive to very many observers, but even so it is surprising to find no reference either to R. J. A. Berry's great work on the architecture of the cortex nor to Wood Jones and Porteous on the 'matrix of the mind'.

Ulcers of the Legs. By P. PIULACHS, Chief Professor Surgeon in the Faculty of Medicine of Barcelona. 10 × 7 in. Pp. 574 + xxvi, with 303 illustrations. 1956. Springfield, Ill.: Charles C. Thomas. (Oxford: Blackwell Scientific Publications.) £5 17s. 6d.

THIS comprehensive work by a well-known Spanish surgeon covers the subject of ulcers of the leg in a very adequate manner. Professor Rudolph Matas, of New Orleans, has written an excellent introduction in which he extols the book in no uncertain manner.

The author has had a very extensive experience of ulcers of the leg, and from this material he has been able to differentiate between vascular and neurotropic ulcers and makes a distinction between ulceration that heals in a short time and the true ulcer which is characterized by its torpidity and stubbornness. The illustrations, which are numerous, are of high quality throughout the book.

One is surprised to find that Unna's paste and plaster is still advocated in the treatment of varicose ulcer; surely the modern medicated elastic bandage is preferable.

The bone changes which follow chronic ulcers of the leg, especially those on the medial side, have been described in minute detail and the histological photomicrographs are excellent.

One of the most interesting chapters in the book is devoted to the complications of venous ulcers which include club-foot, erysipelas, lymphangitis, and cancer.

This is one of the most comprehensive books on the subject of ulcers of the leg yet written and will interest the general, orthopædic, and plastic surgeon.

Subarachnoid Hæmorrhage. By JOHN N. WALTON, M.D. (Durh.), M.R.C.P., First Assistant in Neurology, King's College, University of Durham, in the Royal Victoria Infirmary, Newcastle upon Tyne. With a Foreword by Sir CHARLES SYMONDS, K.B.E., C.B. 8½ × 5½ in. Pp. 350 + xvi, with 25 illustrations as plates. 1956. Edinburgh and London: E. & S. Livingstone Ltd. 30s.

ALTHOUGH subarachnoid hæmorrhage has been recognized since the beginning of this century, it is only during the last decade that real advance has been made in diagnosis and treatment.

Over the years much literature has accumulated dealing with this interesting clinical condition yet this material has not been collected into a single book.

Dr. Walton, who wrote his M.D. thesis on this subject in 1952 for the University of Durham, has done the medical profession of this country a great service by writing this volume.

As Sir Charles Symonds, who writes a foreword to this book, says: "Dr. Walton's survey of his subject is

both comprehensive and detailed. I do not think anything of importance has been omitted." This is real praise for a book, and it is justly deserved.

It will remain the book of reference not only for the neurologist, but also for the physician and the neurosurgeon.

Pott's Paraplegia. By D. Ll. Griffiths, Manchester, H. J. Seddon, London, and R. Roaf, Liverpool. 9½ × 6½ in. Pp. 129 + xiv, with 30 plates and 26 illustrations in the text. 1956. London: Oxford University Press. 50s.

This is a most valuable monograph because it gives the long-term results of three surgeons who have all come to the same conclusions regarding the treatment of Pott's paraplegia. Although tuberculosis of the spine is practically under control in this country, yet it is still a scourge in Asia, Africa, and in some parts of South America.

The authors have written a well-documented book which not only sets out the natural history, clinical features, radiographic features, and pathology of Pott's paraplegia, but also assesses the various forms of treatment. They have come to the conclusion that the best form of operative procedure is anterolateral decompression of the spine for this form of paraplegia; and they give detailed observations on fifty consecutive cases in which this operation was carried out.

The modern antibiotics have not replaced operative treatment in cases of Pott's paraplegia.

This book will receive a world-wide acceptance by the medical profession and will long remain the reference book concerning this important condition.

We could do with more monographs of this calibre in this country.

Cancer of the Lung. Pathology, Diagnosis and Treatment. By Milton B. Rosenblatt, M.D., Associate Professor of Medicine, New York Medical College; and James R. Lisa, M.D., Director, Pathology Service, New York City Department of Hospitals; with J. Maxwell Chamberlain, M.D., Charles L. Daniels, M.D., Harold F. Dorn, Ph.D., N. Chandler Foot, M.D., Chevalier L. Jackson, M.D., George N. Papanicolaou, M.D., Bernard Roswitt, M.D., and Angelo M. Sala, M.D. 9¼ × 6 in. Pp. 330 + xiii, illustrated. 1956. New York: Oxford University Press. (London: Geoffrey Cumberlege.) 120s.

Cancer of the lung is an important problem to the whole of the medical profession, for in the last twenty years there has been an enormous rise in the number of cases seen and reported. It is stated, and quite correctly, that since 1930 the deaths from bronchogenic carcinoma have increased 500 per cent. This being so, such books as this one edited by Rosenblatt and Lisa are of real importance to the medical profession. The authors have enlisted the help of eight contributors, each of whom is a specialist in his own particular field. The book, therefore, is an authoritative work covering all aspects of bronchogenic cancer.

For the specialist there are chapters written by such authorities as Dr. Chevalier L. Jackson on Bronchoscopy; Dr. George N. Papanicolaou and Dr. N. Chandler Foot on Exfoliative Cytology; Dr. J. Maxwell Chamberlain and Dr. Charles F. Daniels on Surgical Therapy; and Dr. Bernard Roswit on Palliative Therapy.

The surgical section is well written and is full of useful operative techniques. It is not sufficiently well known amongst the profession that in this field resectional surgery, in general, may be performed with a mortality rate as low as that for major abdominal surgery.

The whole book is well illustrated and the radiographs are of excellent quality; and the usefulness of the work will interest the pathologist, clinician, radiologist, and surgeon.

La Chirurgie pelvienne après Cloisonnement abdomino-pelvien. Cancer utérin, rectal, vésical. By Henri Bompart and R. Michel-Béchet. With a Foreword by Professor A. Brunschwig. 9¾ × 6¼ in. Pp. 82, with 15 illustrations. 1956. Paris: Masson et Cie. 750 fr.

This small volume describes a new method of utilizing the lower anterior parietal peritoneum to form an anteroposterior flap, which when stitched to the posterior parietal peritoneum at the level of the sacral promontory forms a complete and solid barrier between the contents of the abdomen and the pelvis minor. Such a barrier is indicated in excisions of the rectum, uterus, and bladder. And it is best created at the very beginning of the operation of excision of the organs. It thus allows the excisional or main part of the operation to be carried out unhindered by loops of intestine. But, above all, such a barrier gives the surgeon complete freedom to excise *en masse*, with the organ concerned, the whole or major part of the pelvic peritoneum with its subjacent fat and glands from the pelvic brim downwards.

The book deals fully with all details of this operation. It is well produced and illustrated, and should be read by all abdominal surgeons and gynæcologists. A commendable feature, which has been noted in some other French surgical books published recently, is a short summary in English at the end of the book. The captions, too, are in both English and French.

Chirurgie de la Main. By Marc Iselin, Surgeon to the Maison de Nanterre. Consulting Surgeon to the Hôpital Américain. Second edition. 9½ × 6½ in. Pp. 626, with 319 illustrations. 1955. Paris: Masson et Cie. 4,000 fr.; cloth, 4,800 fr.

Fifty years after the publication of Kanavel's first paper on infections of the hand, we find 986 pages devoted to the surgery of this part. Dr. Iselin's work on surgery of the hand is already very well known; and his personal experience is now embodied in new editions of the two volumes of his book. The first volume, *Livre du Practicien*, deals with acute trauma and infections; and an earlier edition of this was translated into English in 1940.

It is the second volume, *Livre du Chirurgien*, which is the subject of this review. The greater part (two-thirds) of this volume deals with reparative surgery of the sequelæ of trauma and infections of the hand, and the second with congenital and acquired deformities. The hand is the meeting-place of many specialties; and any one surgeon must be endowed with especial qualities to cover the whole subject adequately. It can be said that Iselin has accomplished this task adequately and competently. He gives a detailed and balanced and up-to-date account of the whole subject. Two special chapters are outstanding: in Chapter VI he gives an excellent and fascinating review of the problem of tendon healing and repair; and in Chapter IX the history and development of thumb replacement is particularly good and detailed.

The book is profusely illustrated and well produced. The references given are truly international, up-to-date, and accurate. It is to be recommended as a book of reference and a manual for all surgeons dealing with the hand.

BOOK NOTICES

[The Editorial Committee acknowledge with thanks the receipt of the following volumes. A selection will be made from these for review, precedence being given to new books and to those having the greatest interest to our readers.]

The Invisible Flag. A Report by PETER BAMM, translated from the German by FRANK HERRMANN. 8½ × 5⅝ in. Pp. 229. 1956. London: Faber & Faber Ltd. 18s.

Radiology of the Alimentary Tract in Infancy. By ROY ASTLEY, M.B., M.R.C.S., D.M.R., Radiologist, The Children's Hospital, Birmingham (United Birmingham Hospitals); Clinical Lecturer in Radiology and Lecturer in Radiological Anatomy, University of Birmingham. 9 × 5¾ in. Pp. 287 + viii. Illustrated. 1956. London: Edward Arnold (Publishers) Ltd. 50s.

Principles and Methods of Sterilization. By JOHN J. PERKINS, M.S., Director of Research, American Sterilizer Company, Erie, Pennsylvania. 9¾ × 6¾ in. Pp. 340 + xii, with 174 illustrations. 1956. Springfield, Ill.: Charles C. Thomas. (Oxford: Blackwell Scientific Publications.) 60s.

The British Encyclopaedia of Medical Practice: Medical Progress, 1956. Editor-in-Chief: Sir HENRY COHEN, M.D., D.Sc., LL.D., F.R.C.P., F.F.R. 9⅝ × 6⅝ in. Pp. 380 + xiv. 1956. London: Butterworth & Co. (Publishers) Ltd. 37s. 6d.

Modern Operative Surgery. Edited by the late G. GREY TURNER and LAMBERT CHARLES ROGERS, Professor of Surgery, University of Wales. With a Foreword by Sir GORDON GORDON-TAYLOR, K.B.E. Fourth edition. In two volumes. 9 × 6⅛ in. Vol. II. Pp. 1231–2614, with 614 illustrations. 1956. London: Cassell & Co. Ltd. 75s.

The Neurological Alleviation of Parkinsonism. By IRVING S. COOPER, M.D., Ph.D., F.A.C.S., Director, Department of Neurosurgery, St. Barnabas Hospital, New York; etc. 11 × 8½ in. Pp. 104 + xii, with 68 illustrations. 1956. Springfield, Ill.: Charles C. Thomas. (Oxford: Blackwell Scientific Publications.) 63s.

Cancer of the Esophagus and the Stomach. By OWEN H. WANGENSTEEN, M.D., Minneapolis. Second edition. 9⅜ × 6¼ in. Pp. 144, with 60 illustrations. 1956. New York: American Cancer Society.

Cancer of the Lung. An Evaluation of the Problem. Proceedings of the Scientific Session, Annual Meeting, American Cancer Society, Inc., Nov. 3–4, 1953. 10¼ × 8 in. Pp. 322 + vi, with a number of illustrations. 1956. New York: American Cancer Society.

Boletin Médico del Hospital Infantil, Ciudad de Mexico. Vol. XIII, No. 1–2, Jan.-April. Special numbers dedicated to Dr. Federico Gomez to mark the thirty-fifth anniversary of his graduation from medical school. 9 × 6⅝ in. Pp. 691. Illustrated.

Urology. Edited by MEREDITH CAMPBELL, M.S., M.D., F.A.C.S., New York. With the collaboration of 51 contributors. In three volumes. 9¾ × 6¾ in. Pp. 2356 + complete index in each volume. 1148 illustrations. 1954. Philadelphia and London: W. B. Saunders Co. £15.

Psychosomatic Aspects of Surgery. Edited by ALFRED J. CANTOR, M.D., and ARTHUR N. FOXE, M.D. The Proceedings of the First Annual Meeting of the Academy of Psychosomatic Medicine, held in New York City, October, 1954. 8½ × 5½ in. Pp. 220 + xii. 1956. New York and London: Grune & Stratton Inc. $6.50.

Surgery for Nurses. By JAMES MORONEY, M.B., Ch.B., F.R.C.S. (Eng.), L.R.C.P. (Lond.), Visiting Consultant Surgeon, Clatterbridge General Hospital, Cheshire, and Broadgreen Hospital, Liverpool, etc. With a Foreword by Miss DOROTHY M. SMITH, O.B.E., Chairman of the General Nursing Council for England and Wales. Fourth edition. 8½ × 5½ in. Pp. 703 + xii, with 593 illustrations. 1956. Edinburgh and London: E. & S. Livingstone Ltd. 27s. 6d.

The Year Book of Orthopedics and Traumatic Surgery, 1955-1956. Edited by EDWARD L. COMPERE, M.D., F.A.C.S., F.I.C.S., Professor and Chairman, Department of Orthopedic Surgery, Northwestern University Medical School; etc. 7½ × 5⅛ in. Pp. 333, with 211 illustrations. 1955. Chicago: Year Book Publishers Inc. (London: Interscience Publishers Ltd.) 49s.

The Year Book of Neurology, Psychiatry, and Neurosurgery, 1955-1956. Edited by ROLAND P. MACKAY, M.D. (Neurology); S. BERNARD WORTIS, M.D. (Psychiatry); PERCIVAL BAILEY, M.D., and OSCAR SUGAR, M.D. (Neurosurgery). 7⅝ × 5⅛ in. Pp. 576, with 97 illustrations. 1956. Chicago: Year Book Publishers Inc. (London: Interscience Publishers Ltd.) 53s.

Diseases of the Heart and Circulation. By PAUL WOOD, O.B.E., M.D. (Melbourne), F.R.C.P. (Lond.), Director, Institute of Cardiology, London; Physician, National Heart Hospital; Physician in Charge, Cardiac Department, Brompton Hospital. Second edition. 9½ × 6¼ in. Pp. 1005 + xl, with many illustrations. 1956. London: Eyre & Spottiswoode. £5 5s.

Cardiac Pressures and Pulses. A Manual of Right and Left Heart Catheterization. By ALDO A. LUISADA, M.D., Chicago Medical School; and CHI KONG LIU, M.D., Chicago Medical School. 10 × 6⅝ in. Pp. 116 + viii, with 51 illustrations. 1956. New York and London: Grune & Stratton. $6.00.